Urology Case Reports 35 (2021) 101543

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr

Pediatrics

Inverted urothelial papilloma: A rare pathology in young girl. A case report and literature review

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ARTICLE INFO

Keywords:
Bladder
Paediatric
Inverted urothelial papilloma
Urothelial neoplasm

ABSTRACT

Bladder cancer is relatively common in the general population but is considered a rare entity in children. Rhabdomyosarcoma is the most frequently encountered bladder tumor in children. Inverted papilloma of the urinary bladder is a rare presentation in adults and is considered extremely rare in the pediatric age group. We report a case of inverted urothelial papilloma (IUP) in the bladder in an 8-year-old girl who presented with painless gross hematuria. Radiological investigations, cystoscopy, and histological examination revealed the rare pathology of IUP of the bladder and a resection was performed. No recurrence was encountered after 3 years of follow-up.

Introduction

Inverted urothelial papilloma (IUP) was first described as "polypoid adenoma" by Paschkis in 1927, and subsequently renamed by Potts and Hirst as inverted papilloma.[1] The bladder neck and trigone are considered the most common location for an inverted papilloma, and the lesions usually have a benign course.[2] The aim of this study was to report a case of IUP in the bladder of an 8-year-old girl, highlighting the clinical presentation, diagnosis, treatment, and follow up.

Case presentation

An 8-year-old girl, medically free, presented to Prince Sultan Military Medical City (PSMMC) with a recurrent, painless, gross hematuria. She had no history of passive smoking or trauma. A physical examination was normal and her laboratory results were unremarkable. Abdominal and pelvic computed tomography (CT) scans showed a small urinary bladder lesion in the posterior wall adjacent to the left vesicoureteric junction (Fig. 1).

A cystoscopy examination confirmed a papillomatous tumor with a thin stalk on the back wall of the bladder (Fig. 2) near the left ureteric orifice. A complete transurethral resection of the bladder tumor was done, followed by hemostasis. Histopathological examination of the specimen reported IUP (Fig. 3).

Follow-up of the patient over the next 3 years revealed no more episodes of hematuria. Follow-up cystoscopy done twice at 6- and 18-month intervals showed no evidence of tumor recurrence.

Discussion

IUP is a rare tumor entity that accounts for 1–2% of urothelial tumors.[2] Adenourothelioma and Brunnian adenoma were alternatives terms for IUP.[3] More than 1000 cases of IUP have been reported in the literature since its first description in 1963 by Potts and Hirst.[4] The peak incidence is between the fifth and sixth decade of life, with a male-to-female ratio of 4:1.[1] IUP mainly affects the bladder (90%), but it can occur in any location throughout the urinary tract. The classical presentation for IUP is hematuria, but it might also cause storage bladder symptoms.[3]

Potts and Hirst proposed that the etiology is due to neoplastic transformation for the basal cells of the subtrigonal Home's glands or the subcervical Albarran's glands. However, this explanation is very unlikely because IUPs were later reported outside the bladder.[4] Cummings and Matz et al. assumed that IUP is not truly neoplastic, but instead

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https://doi.org/10.1016/j.eucr.2020.101543
Received 5 December 2020; Received in revised form 15 December 2020; Accepted 17 December 2020
Available online 19 December 2020

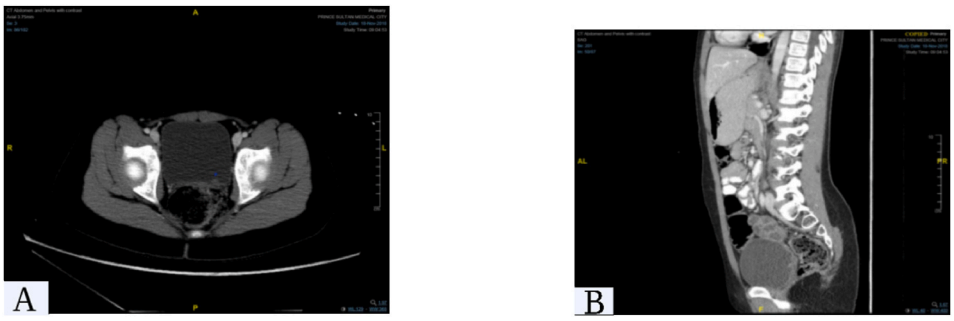

Fig. 1. Abdominal and pelvic CT scans showed small urinary bladder lesion in the posterior wall adjacent to the left vesicoureteric junction. – (1A) Axial view (1B) sagittal view.

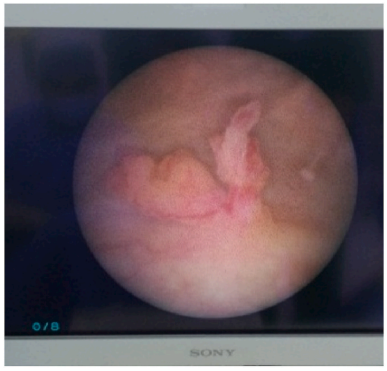

Fig. 2. Cystoscopy examination showed papillomatous tumor with a thin stalk on the back wall of the bladder near left ureteric orifice.

represents a hyperplastic reaction, especially in cases of Brunn's cell nests due to irritative agents or chronic inflammation. This assumption is supported by the predominance of IUP in areas of greatest irritative potential and the low recurrence rate of IUP.[4] Currently, most scientists consider IUP to be a true neoplasm in the urinary tract, even though the precise tissue of origin and the causative agents or processes are still not clear.[4]

Other benign urinary bladder lesions can mimic IUP; these include nephrogenic adenoma and inflammatory pseudo tumor (pseudosarcomatous fibromyxoid tumor), as both these tumors show a bladder mass on ultrasound (US) or a filling defect on intravenous urography (IVU).[5] Cystoscopy can be used for further differentiation of inflammatory pseudotumors, which can be characterized by the gross appearance of inflammation or ulcerative areas, as IUPs typically appear pedunculated or polyploid.[5]

The clinical significance of IUP remains debatable. Until the 1970s, IUP was regarded as a benign tumor that affected the epithelium of the urinary tract and mainly the bladder, However, in the 1980s, several reported cases showed that IUP has malignant potential, based on evidence of malignancy, recurrences, or recurrence with urothelial carcinoma.[4]

Follow-up and management protocols regarding this category of bladder tumor are not well structured, and debate continues regarding the nature of the disease and the potential for malignant transformation. However, the gold standard regarding this lesion is complete resection following the european association of urology (EAU) guideline, followed by close surveillance later on. The intervals for follow-ups and radiation exposure are not clear, but early short-interval follow-ups might be the safest option. Our short-term follow-up revealed no recurrence.

Conclusion

Inverted papilloma of the bladder in children is an extremely rare pathology. Complete tumor resection and early, close, short-term follow-up seems to be a safe treatment option. Additional studies with larger numbers of cases, as well as establishing guidelines for this pathology, will be helpful for physicians and beneficial for patients with IUP.

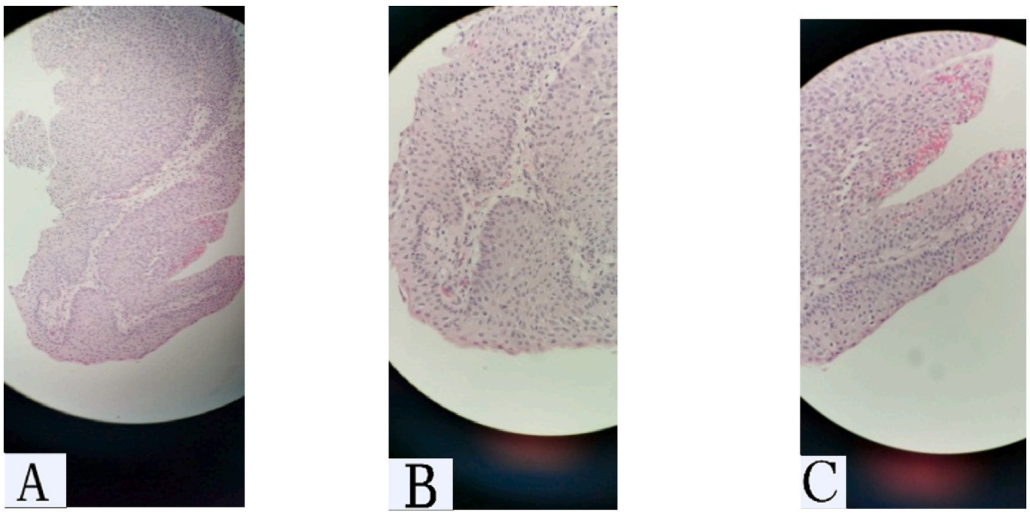

Fig. 3. Histopathological examination of the specimen confirmed Inverted urothelial papilloma.

Consent form

A written consent was obtained from the patient for publication of this case report and accompanying images.

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