

Inverted papilloma of the urinary bladder: A rare cause of haematuria in a young girl

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ABSTRACT

Inverted papilloma of the urinary bladder is rare. In the paediatric group, this condition is even more rare. To date only five cases have been reported in the English literature. Due to its rarity, the morphology and biological behaviour are still uncertain. We report a case of inverted papilloma of the urinary bladder in a 12-year-old girl who presented with recurrent haematuria. The patient's symptoms resolved after endoscopic resection with no recurrence on cystoscopy surveillance for last two years. The magnetic resonance imaging features of this lesion are described.

Keywords: Urinary bladder tumour, neoplasm, inverted papilloma, endoscopic resection

INTRODUCTION

Inverted papilloma of urinary tract was recognised as a benign tumour of urothelium. This lesion was first described by Potts and Hirst in 1963.¹ To date, more than 322 cases of inverted papilloma were reported to involve urinary tract.² They noted that this lesion had potential of recurrent (3.85%) and related to subsequent transitional cell carcinoma (TCC)(1.54%).² Out of those cases, only five cases involved pediatric age group.² In view of extremely rare incidence in paediatric age group the morphology as well as biological behaviour was still not clear. We report an

additional case of inverted papilloma in a paediatric patient and discussed the imaging morphology of the lesion.

CASE REPORT

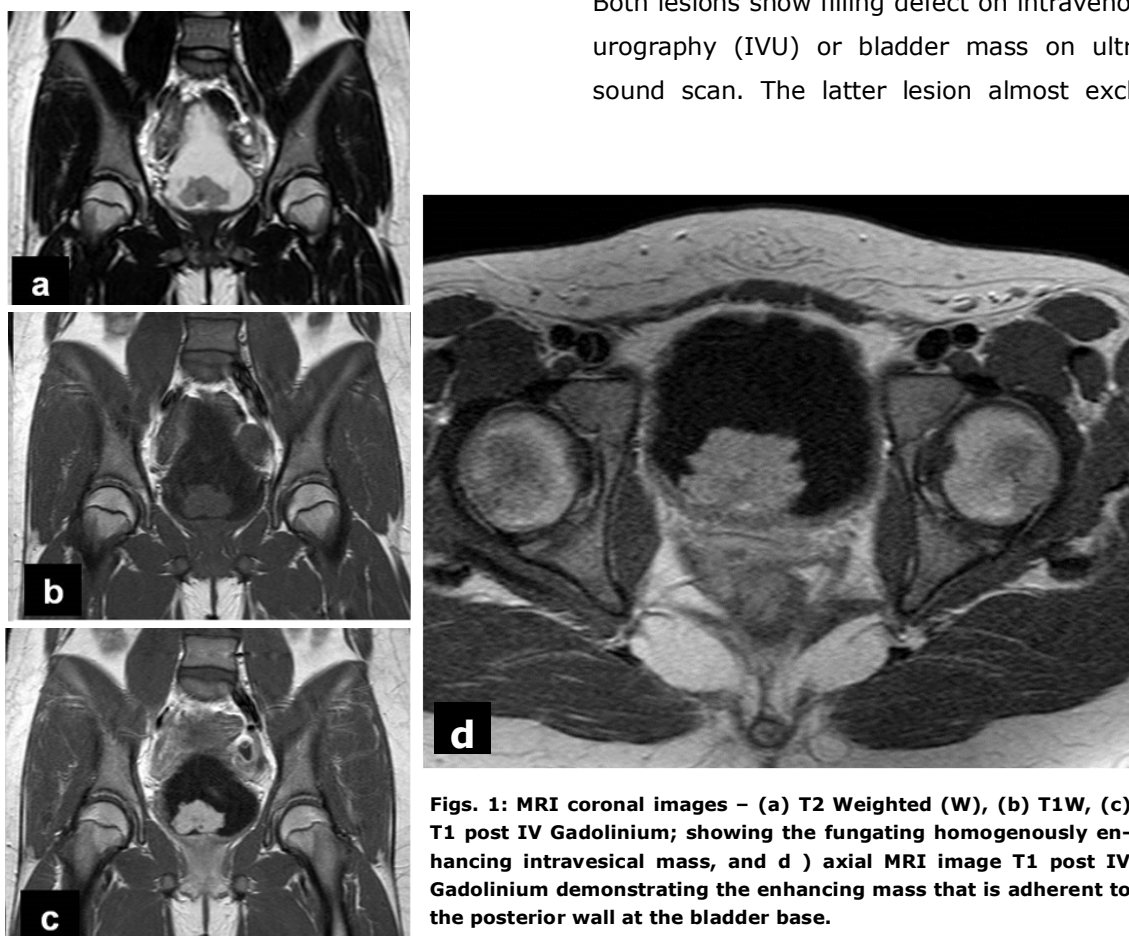
A 12-year-old girl was referred recurrent painless haematuria of one year duration. She claimed that it was continuous haematuria with occasionally mixed with small blood clots. Besides that she denies experience of other urology symptoms such dysuria, increase frequency, nocturia or urinary retention. There were also no anemic symptoms. Physical examination and hematological work up were normal. The initial ultrasound showed presence of bladder mass, and the managing team (paediatrician) proceeded with magnetic resonance imaging (MRI) which showed a fungat-

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ing mass arising from the right lateral wall, measuring approximately 3.7 x 2.6 x 2.3cm. The lesion was hypointense on T2, but slightly hyperintense on T1 imaging, with homogeneous enhancement post intravenous Gadolinium (Figures 1a-d). We proceeded with examination under anaesthesia (EUA) and endoscopic evaluation. We found a four cm mucosal lesion located lateral to the right ureteric orifice. The growth was completely resected endoscopically. Histopathology revealed the lesion to be an inverted papilloma of the urinary bladder (Figure 2). She had an uneventful recovery and she was discharged on three days after surgery. Subsequently, she underwent regular three monthly cystoscopy surveillance without any evidence of recurrence in the last two years.

DISCUSSION

Inverted papilloma accounts for less than 1% of all urothelial tumours with majority reported in adult. It has a male preponderance with ratio of 4:1 and a peak incidence in the sixth and seventh decade of life.² So far, only five cases in children have been reported in the English literature and one case reported in girl.³⁻⁵ Since it is rare in children, the morphology and its biological behaviour are still unclear. In addition to this, the clinical morphology and radiological finding of benign bladder growths are usually not specific, and may overlap between congenital inflammatory, idiopathic or infectious masses.⁶ Among other benign urinary bladder growths, inflammatory pseudo tumour (pseudosarcomatous fibromyxoid tumour) and nephrogenic adenoma clinically may mimic inverted papilloma. Both lesions show filling defect on intravenous urography (IVU) or bladder mass on ultrasound scan. The latter lesion almost exclu-



Figs. 1: MRI coronal images – (a) T2 Weighted (W), (b) T1W, (c) T1 post IV Gadolinium; showing the fungating homogeneously enhancing intravesical mass, and d) axial MRI image T1 post IV Gadolinium demonstrating the enhancing mass that is adherent to the posterior wall at the bladder base.

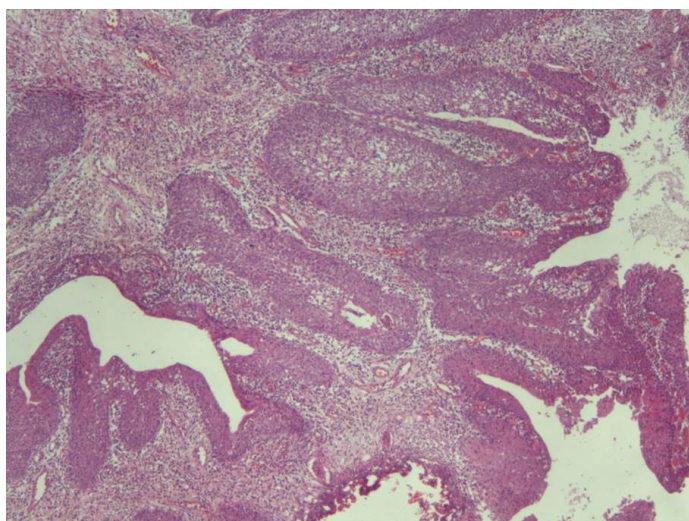


Fig. 2: Endophytic growth of epithelium invaginate into the underlying lamina propria. No nuclear atypia was seen. (H&E stain, x10).

sively occur in adult, thus not a main differential to consider in a young patient. On MRI imaging, inflammatory pseudotumour appears hyperintense on both T1 and T2 weighted images, whereas in our case the lesion appeared hypointense on T2. Both lesions enhance on gadolinium contrast.⁶ Further differentiation between these two lesions can be made during cystoscopy. Gross appearance of inflammation or ulcerative areas are commonly noted in inflammatory pseudotumours, whereas inverted papillomas typically appear polypoidly or pedunculated, as in our case.

The mainstays of management for this lesion are examination under anaesthesia (EUA), complete resection and surveillance cystoscopy. In clinical practice, detailed imaging is rarely done except for staging purposes, if there is concern or evidence of malignant transformation.⁴ Thus, a majority of the previous reported cases had been managed with endoscopic resection after EUA without contrasted radiological study. In our case, MRI performed prior to surgery gave us the opportunity to study the MRI features of an inverted papilloma. However, study based on one

patient may not give clear pictures of all variety of inverted papilloma. Further study with larger sample size is required before reaching the consensus of special imaging features for this lesion.

The gold standard treatment for benign urinary bladder lesions are complete resection and surveillance cystoscopy. However, most authors regard inverted papilloma as neoplasm with malignant potential.^{7, 8} This was supported by few reported cases of transformation to malignant papilloma in adult patient.⁸ Therefore, strict surveillance with cystoscopy is mandatory for all cases. Recently this practice has been challenged.⁹ One study showed no recurrence was noted in 24 patients who were followed up for a mean duration of 25.8 months (range, 6-58 months).⁹ The authors suggested that rigorous follow up protocol is not required. Another study suggested that the initial histology including urine cytology as a selection for vigorous follow up cases.¹⁰ They considered a single lesion with classical inverted papilloma histology as having benign biological behaviour, and does not need regular surveillance.¹⁰ In patients with

lesion showing other histological type or the presence of atypical cell in urine cytology, they recommended standard surveillance protocol to be followed.¹⁰

In pediatric cases, the interval of surveillance remains debated. In view of its rarity, there is no consensus on interval of surveillance in this group of patient. Most of the authors manage it as similar to adult urothelial carcinoma. To date, none of the five reported cases in paediatric patients had recurrence after four years follow up. Similarly in our case, surveillance cystoscopy showed no recurrence for the last two years. More cases need to be studied and published before we can solve out the standard protocol of management.

In conclusion, we report a rare cause of haematuria in a young patient and we hope this added case does benefit towards that and more cases of paediatric inverted papilloma will be published thereafter.

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