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Case Report

Low-grade papillary urothelial neoplasm: A case report study ☆

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ABSTRACT

Urothelial carcinoma (UC) of the bladder is a prevalent malignant tumor among the elderly, whereas its incidence is scarce in the first 2 decades of life. The most commonly reported symptom in the literature is isolated hematuria, frequently overlooked during the initial medical assessment. In this study, we present the case of a 3-year-old male with hematuria, accompanied by other irritative symptoms such as flank pain, nausea, and vomiting. Ultrasonography revealed a bladder mass, which was later confirmed to be a noninvasive low-grade papillary urothelial carcinoma (NLPUC) through histopathological examination. This report discusses the clinical and pathological characteristics of the case and examines current literature on the topic.

Introduction

Urothelial carcinoma (UC) of the bladder is the fifth most prevalent form of cancer in humans, typically manifesting in the sixth or seventh decade of life. However, it is infrequent in children and young adults [1], affecting only 1%-2.4% of the population below 40 years of age and accounting for only 0.1%-0.4% of cases during the initial decades of life [2–4]. The male-to-female ratio in the pediatric population is 2:1 [5]. UC can be classified into various types, with noninvasive low-grade papillary urothelial carcinoma (NLPUC) being the most common subtype, representing 93.4% of pediatric urothelial carcinomas [5]. The most prevalent symptom is isolated painless gross hematuria, present in 75%-80% of patients [5,6], followed by irritative signs such as abdominal or flank pain [5]. Ultrasonography is the preferred initial imaging modality with a sensitivity of 85%-100% [5]. Although computed tomography (CT) and magnetic resonance imaging (MRI) have been utilized, MRI is preferred over CT to avoid radiation exposure in pediatric patients. Cystoscopy with biopsy provides a definitive diagnosis and enables surgical resection. Transurethral resection of the bladder (TURB) is the standard of care for UC treatment, with a low recurrence rate in pediatric patients. The purpose of this study is to report a new case of UC in the pediatric popula-

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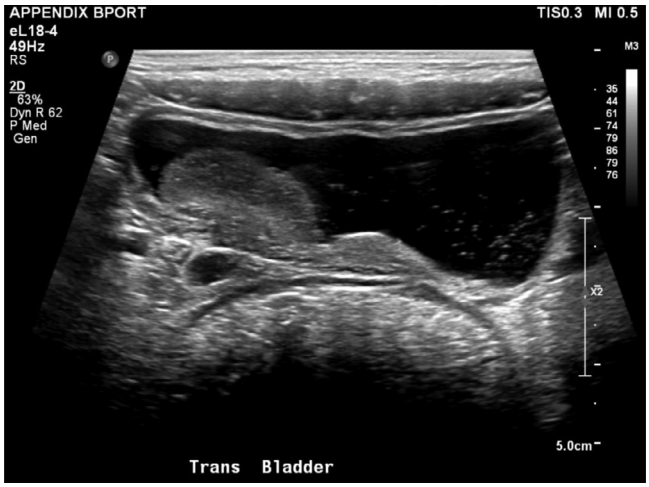

Fig. 1 – Ultrasound of the urinary bladder reveals an iso-echogenic mass on the right side of the bladder wall.

tion, describe its findings, and review recent studies in the literature.

Case report

A 3-year-old male patient was admitted to our hospital with a history of painless hematuria, flank pain, and a fever of 40°C (104°F) for 5 days. An ultrasound examination of the urinary tract revealed a 2.6 × 1.8 × 2.2 cm irregular heterogeneous echogenic mass emerging from the right side of the urinary bladder wall, exhibiting blood flow on color Doppler (Figs. 1 and 2). Additionally, two 4 mm stones were identified at the right kidney's interpolar region/lower pole and the right ureterovesical junction (Figs. 3 and 4), resulting in mild-to-moderate hydroureteronephrosis in the right kidney (Fig. 5). The patient underwent cystoscopy and excisional biopsy, and a right nephrostomy tube was placed to treat the right hydroureteronephrosis. Histopathological examination revealed a papillary configuration of the urothelium with a fibrovascular core (Fig. 6). Moreover, prominent nucleoli and brisk upper-level mitosis were observed, consistent with NLPUC (Fig. 7). The remaining fragments displayed a benign-appearing urothelium with lamina propria edema. The patient could urinate after surgery, and the urologist established a close follow-up plan.

Discussion

This paper presents a case report of a 3-year-old male diagnosed with low-grade papillary urothelial carcinoma (UC) of the bladder, a rare form of cancer in children. The incidence of UC in the first 2 decades of life is reported to be between 0.1% and 0.4% [2–4]. Histologically, UC can be classified into several types, including papillary urothelial neoplasms of low malignant potential (PUNLMP), noninvasive low-grade papillary urothelial carcinoma (NLPUC), and noninvasive high-grade papillary urothelial carcinoma (NHPUC). The low-grade subtype, as found in our case, accounts for 93.4% of pediatric urothelial carcinomas [5].

Clinical manifestations of UC in children include gross hematuria, which is present in 80% of cases [7], as well as other symptoms such as abdominal pain, flank pain, fever, pyelonephritis, nephrolithiasis, recurrent cystitis, and emesis [8]. In some cases, a bladder mass is found incidentally [9]. Risk factors for UC in the pediatric population are not well defined. Although tobacco use is the leading cause of adult UC [8], in a single case report of a 16-year-old boy with UC, smoking history was reported [10], and a case series of 3 UC patients showed a common history of environmental exposure to amines [9]. However, no such history was reported in our patient.

Histopathologically, low-grade papillary urothelial neoplasm is defined as a papillary configuration of the urothelium with a fibrovascular core. Prominent nucleoli and brisk

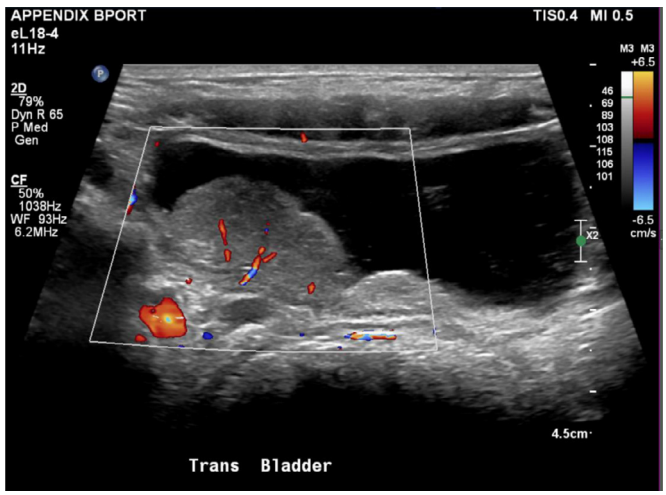

Fig. 2 – Ultrasound doppler of the urinary bladder mass shows blood flow.

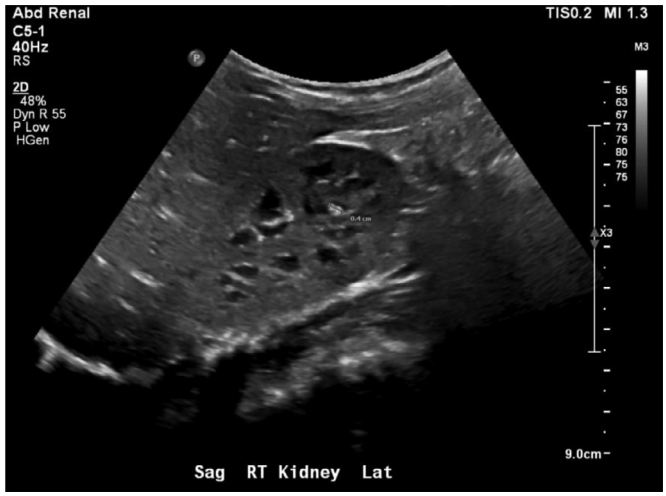

Fig. 3 – Ultrasound of the right kidney shows a 4 mm non-obstructing stone at the interpolar region/lower pole of the right kidney.

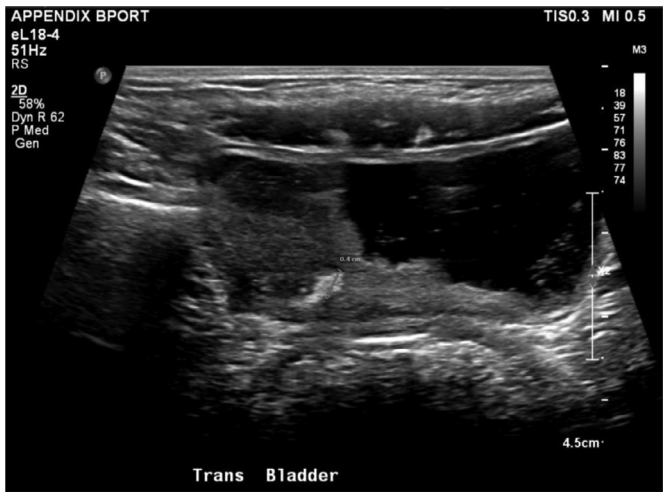

Fig. 4 – Ultrasound of the urinary bladder shows a small stone measuring 4 mm at the right ureterovesical junction.

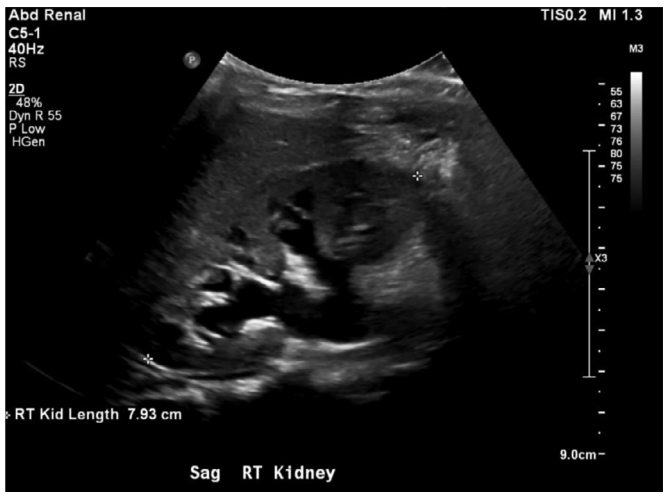

Fig. 5 – Ultrasound of the right kidney depicting mild to moderate hydronephrosis and mild ureteronephrosis in the proximal right ureter.

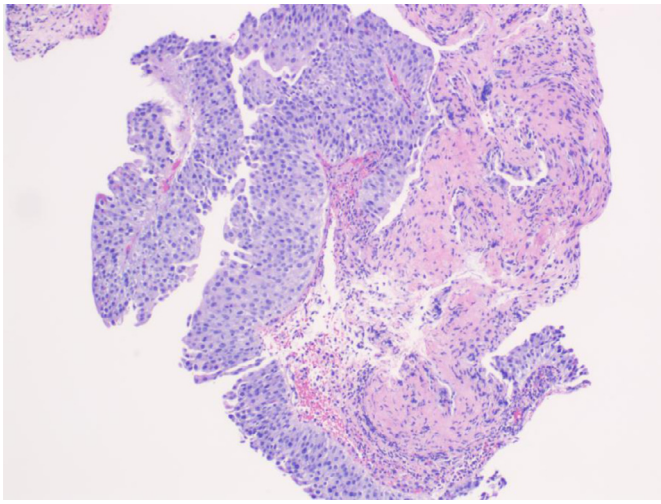

Fig. 6 – A low magnification image depicts the papillary arrangement of urothelium with a fibrovascular core.

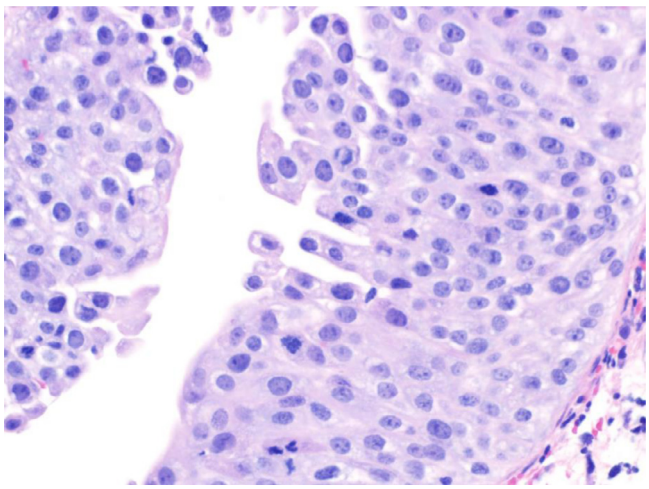

Fig. 7 – A high-magnification view of mitotic figures displays prominent nucleoli and brisk upper-level mitosis, indicating the presence of NLPUC.

upper-level mitosis are present, but there is no invasion through the basement membrane [11].

The optimal diagnostic modality for this condition is still under investigation, with multiple imaging techniques being used in clinical practice. Ultrasound is often the first-line imaging modality due to its widespread availability and lack of ionizing radiation, although its diagnostic performance is operator-dependent [4]. CT is another option, although the potential risks of radiation exposure must be weighed against diagnostic benefits. MRI can be used in selected cases [12]. Cystoscopy is a valuable diagnostic and therapeutic tool that allows for visualization and resection of the mass. Transurethral resection of the bladder (TURB) is the primary treatment modality, with low recurrence rates reported in most studies [13]. In our case, cold cup biopsy forceps were used because of the benign appearance of the lesion and the risk of ureteral injury. Open resection of the lesion is reserved for high-grade lesions.

Currently, there are no established guidelines for the follow-up of pediatric patients with UC. A study suggests that low-grade cases should be followed less frequently 3 years after TURB, as most recurrences occur during the first year [5]. Cystoscopy is the best diagnostic method for detecting recurrence, and it is crucial to establish optimal follow-up strategies for these patients [12]. Although low-grade UC in children has an excellent prognosis and low recurrence rate [1], some studies have reported the possibility of recurrence [14]. Therefore, a nationwide multicenter study is required to include more cases, and further research is needed to determine the optimal follow-up strategy.

Conclusion

In the pediatric and young adult population, gross hematuria is not typically associated with malignancy, but clinicians should consider a wide range of potential differential diagnoses. The prompt use of noninvasive diagnostic modalities, such as ultrasound, is imperative for preventing complications and facilitating accurate diagnosis and treatment. Regarding appropriate follow-up strategies, low-grade urothelial carcinoma (UC) has demonstrated lower recurrence rates after 3 years, suggesting fewer subsequent visits are required for safe monitoring.

Patient consent

The authors declare that the patient's family has provided written consent that encompasses all patient aspects and details, including the use of ultrasound and histology images, in this case report.

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