



Capillary hemangioma of the bladder in a pediatric patient

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ABSTRACT

We report a case of a bladder hemangioma in a pediatric patient. A 2-year-old Caucasian female presented with intermittent gross hematuria and protrusion of beefy red tissue near the vaginal introitus when straining. On cystoscopy, we discovered a wide-based vermiform mass. Transurethral resection of the bladder mass was performed. Based on the histological findings of the tissue resected, a diagnosis of capillary hemangioma of the bladder was made. Despite their rarity, bladder hemangiomas should be considered in the differential in children with gross hematuria.

1. Introduction

Gross hematuria in children is uncommon, accounting for just 0.1% of pediatricians' outpatient visits. The major causes of pediatric gross hematuria include glomerulopathy, urinary tract infections, hypercalciuria, urolithiasis, congenital urinary tract anomalies and trauma. Bladder tumors, however, are among the least common causes. Furthermore, the histologic types of bladder tumors seen in children differ vastly from those observed in adults. The most common pediatric bladder tumor is rhabdomyosarcoma, followed by fibroepithelial polyps.¹ Bladder hemangiomas, however, are exceedingly rare in both adults and children, and therefore relatively few reports of them exist in the literature.^{1,2} Although there is a known association with cutaneous vascular lesions and vascular malformation syndromes (namely, Sturge-Weber syndrome and Klippel-Trenaunay-Weber syndrome), bladder hemangiomas may also occur in the absence of these conditions.³ Here, we present a case of histologically proven hemangioma of the urinary bladder in an otherwise healthy 2-year-old Caucasian female.

2. Case presentation

The patient presented with a one-year history of intermittent gross hematuria. In addition, her parents described occasionally seeing beefy

red tissue protruding near the vaginal entrance often associated with difficulty initiating her urine stream. She was initially treated by a referring provider for urethral prolapse but was referred to pediatric urology when her symptoms failed to resolve.

Initial physical exam was unremarkable. Patient did not demonstrate any distress or urinary posturing and she was found to have normal Tanner stage 1 female external genitalia with no evidence of prolapse or traumatic injury. No lesions consistent in appearance with hemangioma were observed on her skin.

Based on the clinical history, we first obtained a Renal and Bladder Ultrasound (RBUS) to rule out prolapsing ureterocele, fibroepithelial polyp, or other mass of the bladder or urethra that could account for her symptoms. RBUS demonstrated an isoechoic exophytic mass extended into the bladder lumen from the right posterior lateral aspect with associated internal vascularity on Doppler flow (Fig. 1) with possible extension through the bladder wall. The formal radiographic impression was that the bladder mass could represent a fibroepithelial polyp, although rhabdomyosarcoma could not be excluded. Accordingly, we recommended taking the patient to the operating room (OR) later that week for cystoscopy and transurethral resection of bladder mass.

Cystoscopic survey of the bladder demonstrated a vermiform mass extending from the right lateral wall of the bladder near the bladder neck (Fig. 2). It was a long tube-like structure, similar in shape to an appendix and did not have the appearance typical for urothelial

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carcinoma or rhabdomyosarcoma. The base of this tumor was much broader than expected and the worm-like portion widened at the base as it entered the bladder making it far too thick to avulse with cold cup biopsy forceps. A resectoscope and Collins knife were used to transect the tumor at its base. The transect portion of the tumor was grasped and extracted through the urethra. Approximately 1.5–2 cm of tissue was obtained. Frozen section revealed that the mass was likely benign and not consistent with known malignancies. The findings were also discussed peri-operatively with a pediatric oncologist at our center. Ultimately, given the likely benign nature of the mass, we determined that complete excision would be indicated to prevent recurrence. Owing to the limitations of the pediatric resectoscope and the location of mass near the bladder neck, we felt it would be very difficult to complete endoscopically. The parents were informed of our findings peri-operatively and opted to return for an elective wide local excision of the tumor base to be performed at a later date once final pathology results were known.

Pathologic examination subsequently revealed the polypoid tissue to be reactive urothelium superficially with a proliferation of capillary-sized vessels and prominent “feeder vessels” in the deeper portion of the sample (Fig. 3). There was no significant cytologic atypia or mitotic

activity, and immunohistochemical staining was positive for CD31 and desmin, consistent with endothelial cells and vascular smooth muscle, respectively. A diagnosis of capillary hemangioma was made based upon these final pathology results.

The patient was taken back to the OR about 1 month later for open complete excision of the tumor base. She was discharged on post-operative day 1 without a catheter and minimal morbidity. A small amount of residual hemangioma with negative margins was seen on final pathology. The patient is currently asymptomatic on ultrasound surveillance with no evidence of recurrence at 1 year.

3. Discussion

Though the literature on pediatric bladder hemangiomas is rather sparse, it is thought that these most commonly present with intermittent painless macroscopic hematuria. However, they can also present with life threatening complications; at least one account of massive hemorrhage with consequent hypovolemic shock in a toddler with this diagnosis has been described.⁴ Roughly one third of patients with a bladder hemangioma will also have hemangioma(s) in other locations. These

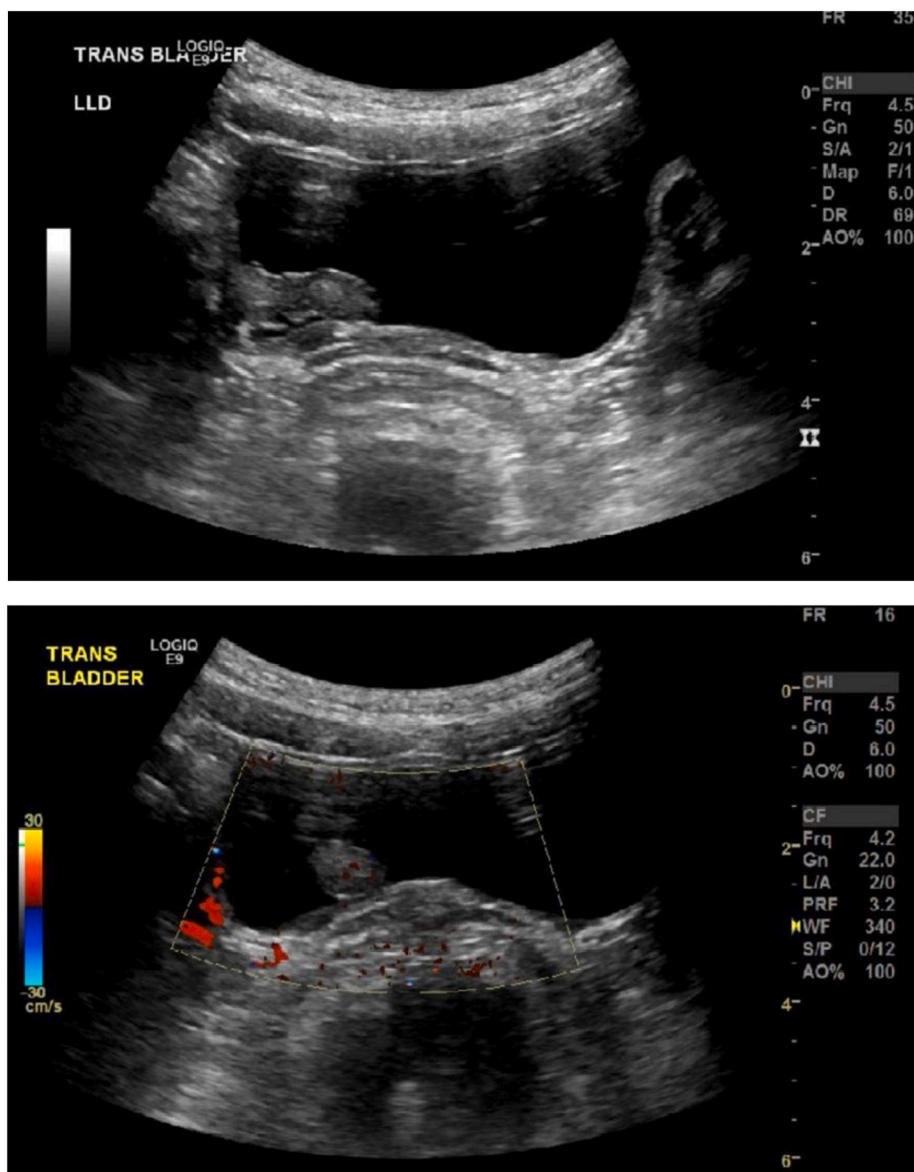


Fig. 1. Transverse views of the bladder on RBUS showed an isoechoic exophytic mass extending into the bladder lumen from the right lateral wall with associated internal vascularity on Doppler flow.

benign tumors are most often found in the dome, along the posterolateral walls, or in the trigone of the bladder, and are usually confined to the submucosa. In most cases, they occur as solitary lesions but multiplicity has been seen on rare occasion in both children and adults. With respect to size, our patient's tumor was rather atypical; Cheng et al.'s review of bladder hemangiomas in 19 adult patients over a 60-year period demonstrated a median size of just 0.7 cm, whereas our

patients was at least double that size in length.⁵ Three histologic subtypes of hemangioma have been described: cavernous, capillary and arteriovenous. Although our patient was found to have the capillary form, the most common of the three is the cavernous subtype. On ultrasound, bladder hemangiomas take on a multitude of appearances: they may be hyperechoic, hypoechoic, or even isoechoic and can appear as a well-margined solid mass or little more than diffuse bladder wall

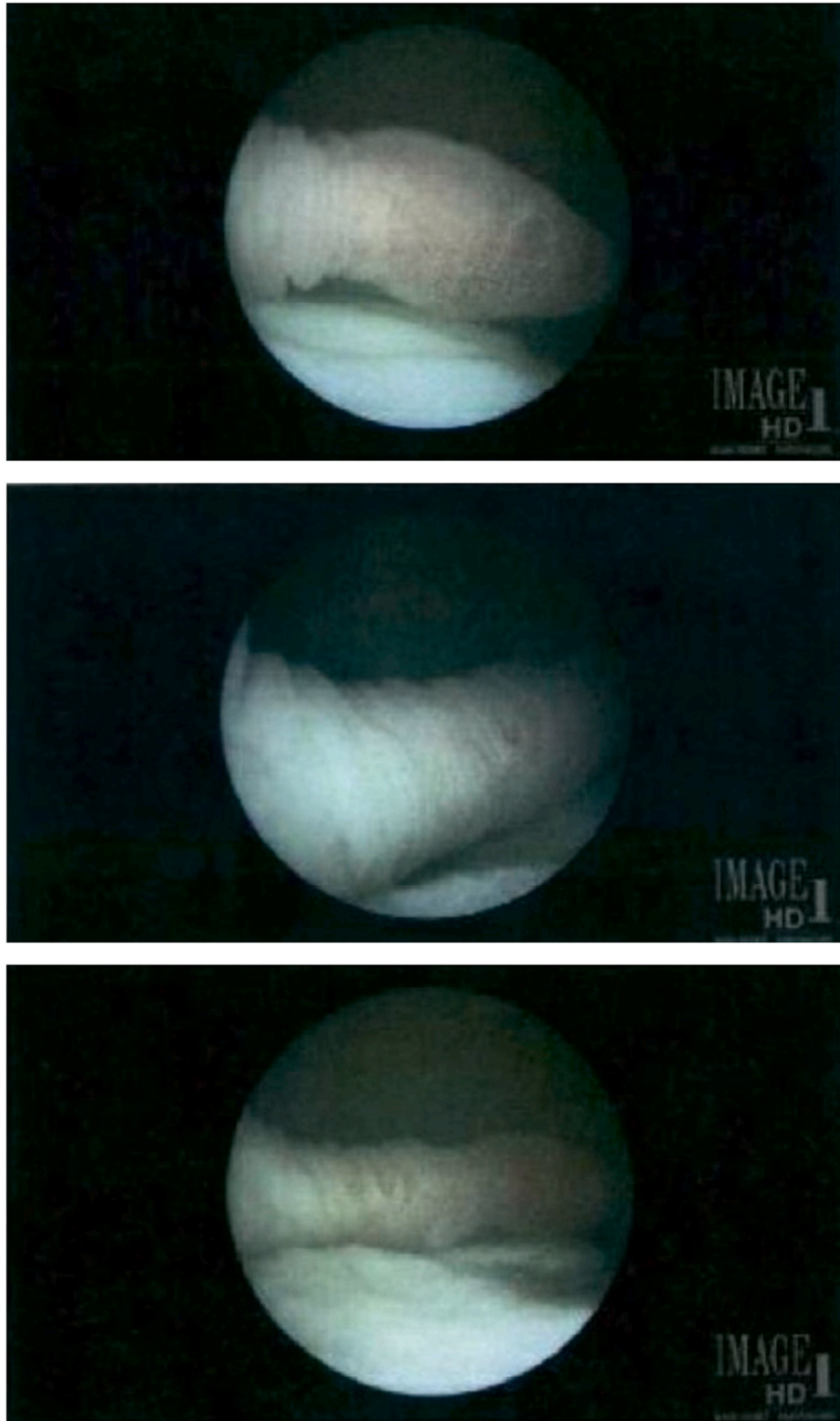


Fig. 2. Vermiform mass extending from the right lateral wall of the bladder near the bladder neck, as seen on cystourethroscopy.

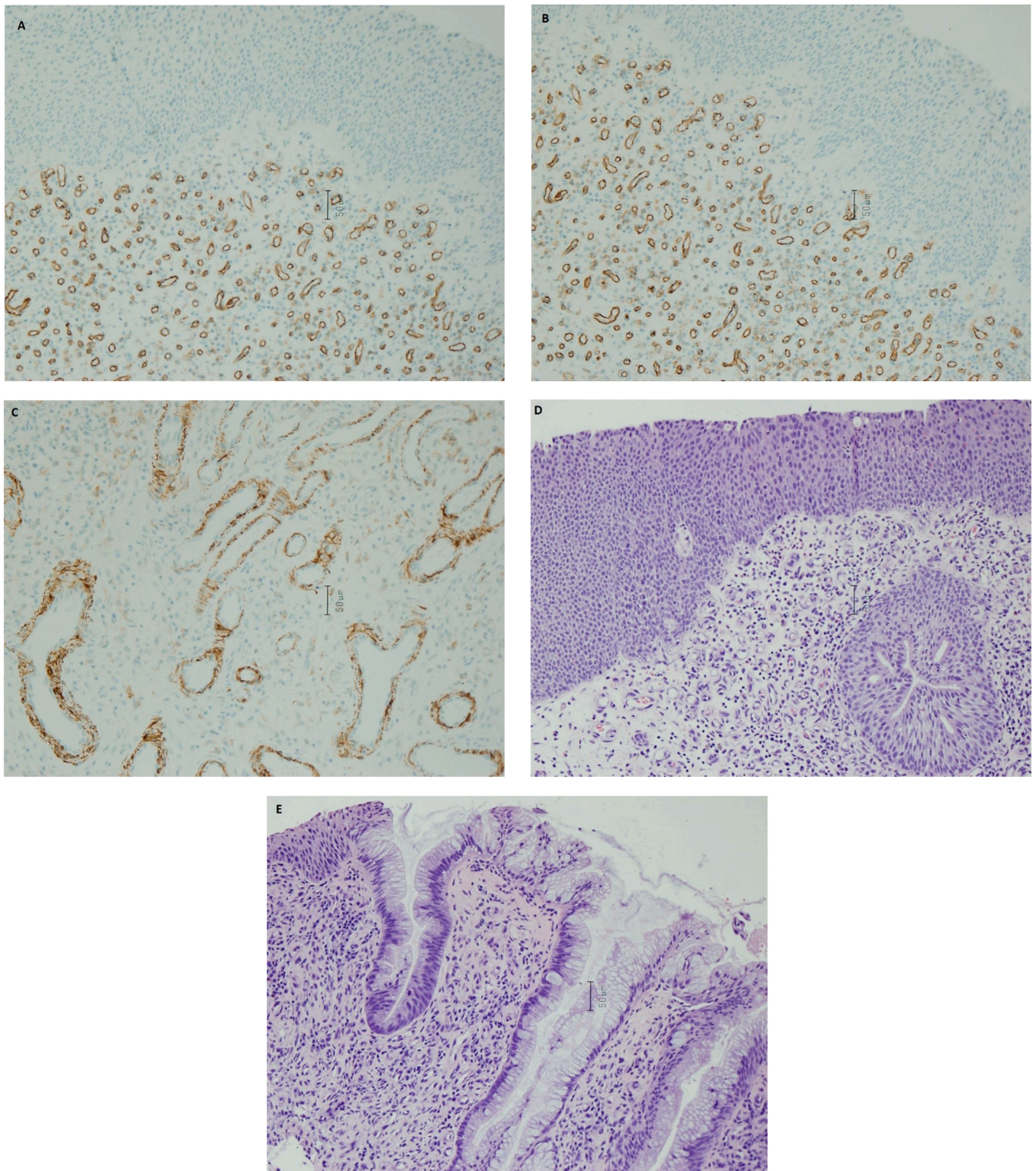


Fig. 3. Histologic appearance of urinary bladder hemangioma. a-b. Immunohistochemical staining for CD31 was positive. c. Desmin stain was positive. d-e. Reactive urothelium with the proliferation of capillary sized vessels and prominent feeder vessels noted in the deeper portions of the tissue sample.

thickening with calcifications.

4. Conclusion

The differential diagnosis of a child with painless gross hematuria, difficulty urinating and a tissue protruding from the introitus includes rhabdomyosarcoma (RMS), urethral mucosa prolapse, and prolapsed

ureterocele. Based on our experience, we conclude that urinary bladder hemangioma should also be considered in this differential diagnosis.

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Consent

Consent was obtained.

Declaration of competing interest

None.

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