

Research Article

An Unusual Cause of Gross Haematuria in a Child: Case Series

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Abstract: **Introduction:** Haemangioma of the urinary bladder is an exceedingly rare benign vascular tumour, accounting for approximately 0.6% of all bladder tumours, and is uncommon in childhood. Because recurrent painless gross haematuria is its most frequent presentation and it has no pathognomonic clinical features, it is easily mistaken for a malignant neoplasm. We report a paediatric case managed successfully by open bladder-sparing excision. **Case presentation:** A 10-year-old boy presented with three days of painless, continuous gross haematuria with clots and obstructive lower urinary tract symptoms, on a background of a similar self-limiting episode at three years of age. He was severely anaemic (haemoglobin 5.4 g/dL). Ultrasonography and contrast-enhanced computed tomography demonstrated a vascular, nodular lesion at the bladder dome with an associated intravesical clot, raising suspicion of a neoplasm. Cystoscopy revealed bluish-red vascular lesions over the dome. Through an extraperitoneal open approach the feeding vessels were ligated and the lesion was widely excised with a rim of normal bladder wall. Histopathology confirmed a cavernous haemangioma with no evidence of malignancy. **Conclusion:** Bladder haemangioma should be considered in the differential diagnosis of recurrent gross haematuria in children. Diagnosis rests on cystoscopy and histopathology, and management is individualised to lesion size and depth. Complete excision with a safe margin is curative and prevents the recurrent, potentially life-threatening haematuria that characterises untreated disease.

Keywords: Bladder haemangioma; Cavernous haemangioma; Gross haematuria; Child; Paediatric urology; Partial cystectomy.

INTRODUCTION

Haemangiomas are benign tumours of vascular origin that may arise at almost any site but are distinctly uncommon in the genitourinary tract. Within the urinary bladder they are exceedingly rare, representing only about 0.6% of all bladder tumours.^{1,2} The lesion is thought to be congenital, arising from embryonic angioblastic stem cells that persist within the bladder wall, which explains its occurrence across all age groups and its recognised association with certain congenital vascular syndromes.¹

Although bladder haemangioma occurs at any age, it is relatively rare in childhood and shows a slight male predominance. Most reported lesions are solitary and of the cavernous subtype, with a predilection for the dome,

posterior wall and trigone, and range in size from a few millimetres to about 10 cm.² The dominant clinical feature is recurrent, isolated, painless gross haematuria, which may be severe enough to cause anaemia or even haemorrhagic shock.³ Because these features are non-specific and imaging frequently suggests a neoplasm, the pre-operative distinction from malignant bladder tumours is difficult, and definitive diagnosis depends on cystoscopy and histopathological confirmation.²

We describe a 10-year-old boy who presented with recurrent gross haematuria and profound anaemia, in whom a vascular dome lesion was excised with a bladder-sparing open technique and confirmed histologically to be a cavernous haemangioma.

Case-presentation

A 10-year-old boy presented with a three-day history of gross haematuria and lower abdominal pain. The haematuria was painless, continuous and associated with the passage of clots and obstructive lower urinary tract symptoms. He had experienced a similar episode at the age of three years, which had been managed conservatively. There was no history of trauma, flank pain or fever, and no significant past medical or family history. Vital signs and systemic examination, including the skin, were unremarkable, with no cutaneous vascular lesions to suggest an associated syndrome.

Investigations

Laboratory evaluation revealed severe anaemia with a haemoglobin of 5.4 g/dL and a haematocrit of 17.4%. The total leucocyte count was 12,800/mm³ and the platelet count 2.17 lakh/mm³ ($217 \times 10^9/L$). Blood urea was 37 mg/dL and serum creatinine 0.7 mg/dL, and the coagulation profile was normal. Urine microscopy showed numerous red blood cells with mild proteinuria and no casts.

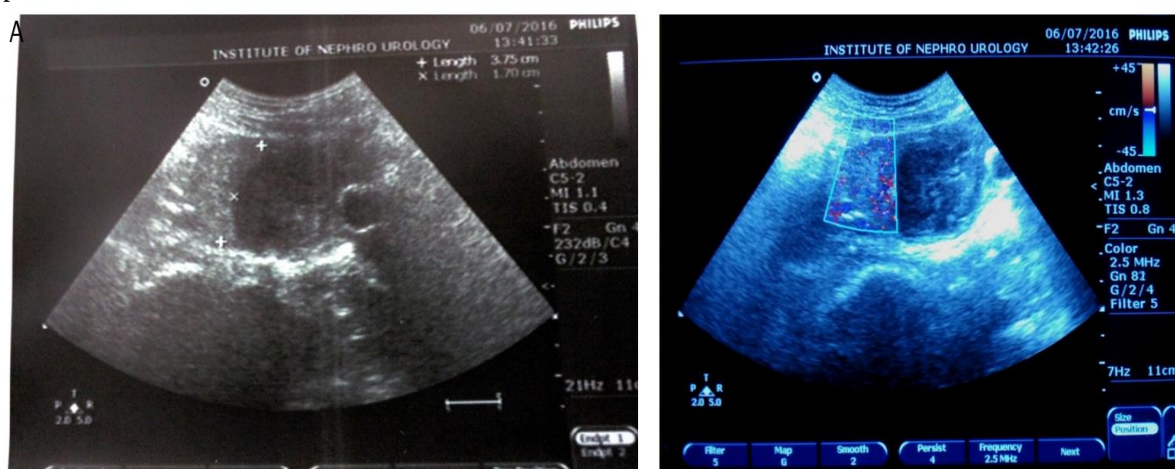


Figure 1. Pelvic ultrasonography of the urinary bladder. (A) Intravesical clot with a hyperechoic area at the bladder dome measuring approximately 3.3×1.7 cm. (B) Colour Doppler interrogation demonstrating mild vascularity within the dome lesion. Both kidneys were normal without hydronephrosis or calculi.

Pelvic ultrasonography revealed a bladder clot together with a hyperechoic area at the dome of the bladder measuring 3.3×1.7 cm, which showed mild vascularity on Doppler interrogation (Figure 1). Both kidneys were grossly normal, with no evidence of hydronephrosis or urinary calculus.

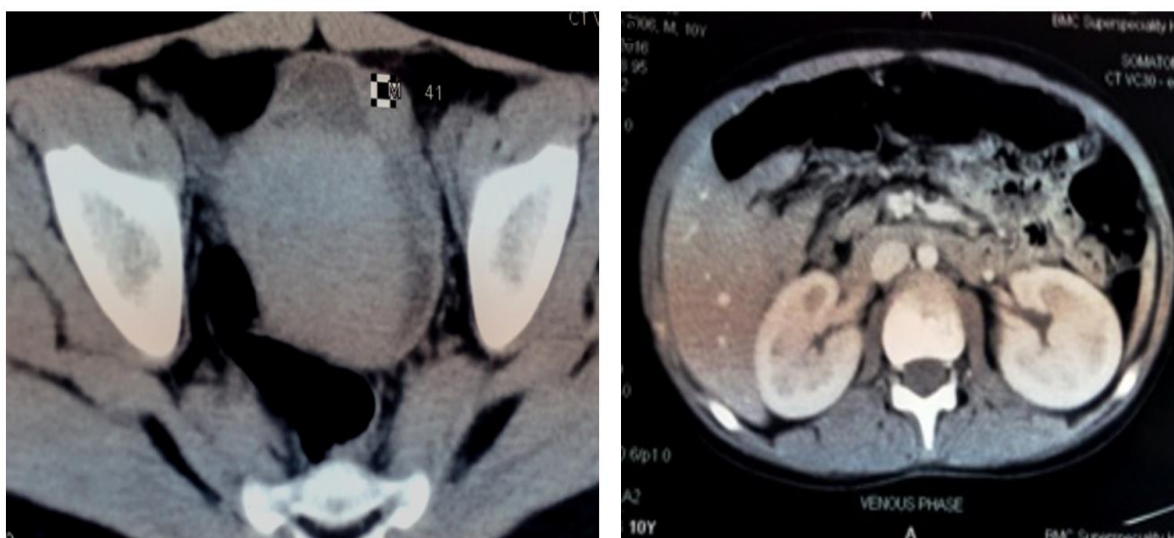


Figure 2. Contrast-enhanced computed tomography (CECT) of the kidneys, ureters and bladder. (A) Nodular wall thickening of the superior aspect of the urinary bladder measuring approximately 4×3 cm, showing minimal enhancement and raising suspicion of a neoplastic growth. (B) A well-defined hyperdense intravesical lesion measuring approximately 5×7 cm, consistent with a bladder clot. Both kidneys were normal.

Contrast-enhanced computed tomography (CECT) of the kidneys, ureters and bladder showed nodular wall thickening of the superior wall of the urinary bladder measuring 4×3 cm, suspicious for a neoplastic growth, together with a well-defined, minimally enhancing hyperdense intravesical lesion measuring 5×7 cm consistent with a bladder clot (Figure 2). Both kidneys were normal.

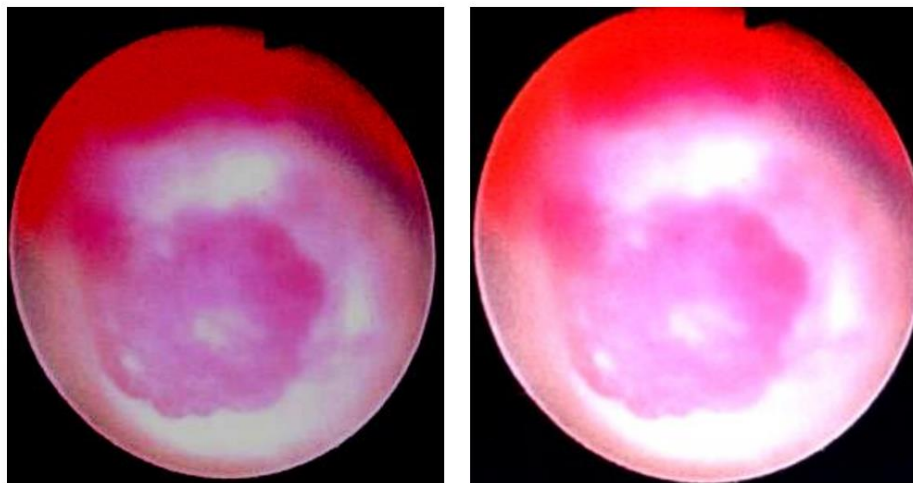


Figure 3. Cystoscopic appearance. (A, B) Bluish-red vascular lesions occupying the bladder dome and measuring approximately 4×3 cm, with an associated overlying clot measuring approximately 4×6 cm.

Cystoscopy revealed bluish-red vascular lesions occupying the bladder dome and measuring 4×3 cm, with an associated bladder clot measuring 4×6 cm (Figure 3). The appearance was characteristic of a vascular lesion rather than a papillary urothelial tumour.

Operative Procedure

After correction of anaemia, the patient was taken up for open surgical exploration. Surgery was performed through a lower midline abdominal incision and the urinary bladder was approached by an extraperitoneal route. On exploration, multiple bluish-red vascular lesions were confirmed over the dome of the bladder, together with an intravesical clot. The feeding vessels were identified and ligated, and a wide excision of the lesion with a rim of normal bladder tissue was carried out. The specimen was sent for histopathological examination and the bladder was closed in layers (Figure 4).

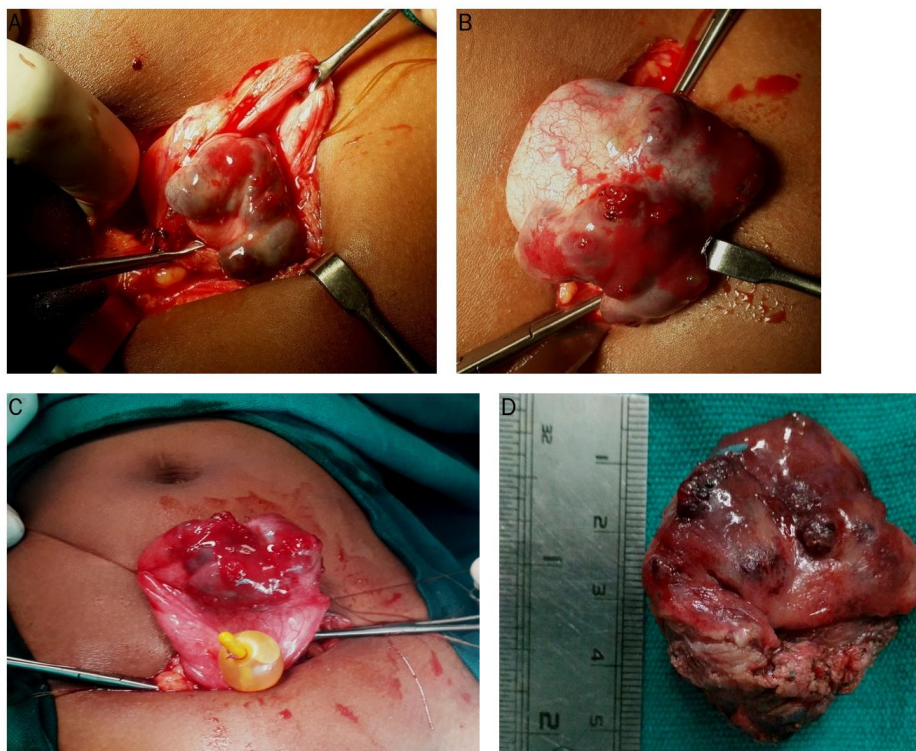


Figure 4. Intraoperative photographs. (A, B) Open extraperitoneal exposure of the urinary bladder showing multiple bluish-red vascular lesions over the dome. (C) Wide excision of the lesion with a rim of macroscopically normal bladder wall after ligation of the feeding vessels. (D) Gross excised specimen.

Histopathology

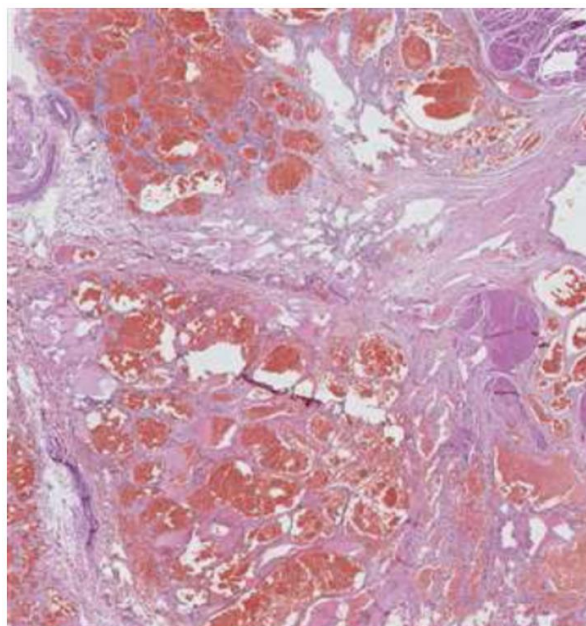


Figure 5. Photomicrograph of the excised lesion (haematoxylin and eosin). The sections show fibromuscular tissue containing dilated and congested blood vessels of varying calibre separated by a fibrovascular stroma, lined by flattened endothelium — features diagnostic of a cavernous haemangioma. No granulomas or malignancy were identified. Microscopic examination showed fibromuscular tissue with dilated and congested blood vessels separated by a fibrovascular stroma. The vascular channels were lined by a flattened endothelium, and the sections were negative for granulomas or malignancy. The findings were diagnostic of a cavernous haemangioma of the urinary bladder (Figure 5).

Outcome and Follow-up

The postoperative course was uneventful and the haematuria resolved completely after surgery. The decision for operative removal in this child had been driven by progressive anaemia and a large vesical mass with radiological suspicion of malignancy. Given the recognised, though rare, associations of bladder haemangioma with cutaneous and systemic vascular syndromes, a systemic evaluation was performed and revealed no additional findings. The patient was advised regular follow-up to detect any recurrence or residual disease.

Case 2. Botryoid Embryonal Rhabdomyosarcoma of the Urinary Bladder

Case Presentation

A 4-year-old boy presented with a two-week history of intermittent gross haematuria accompanied by straining during micturition, a poor urinary stream and two episodes of acute urinary retention. The parents had noticed increasing lower abdominal fullness. The haematuria was painless and occasionally associated with the passage of small friable tissue-like fragments in the urine. There was no history of trauma, fever, weight loss or preceding urinary tract infection, and no relevant family history. On examination the child was afebrile and haemodynamically stable; a firm, non-tender suprapubic mass was palpable. The external genitalia and skin were normal, and there was no lymphadenopathy or hepatosplenomegaly.

Investigations

Laboratory evaluation revealed mild anaemia with a haemoglobin of 9.8 g/dL and a haematocrit of 30.2%. The total leucocyte count was 9,600/mm³ and the platelet count 3.4 lakh/mm³ (340 × 10⁹/L). Blood urea was 28 mg/dL and serum creatinine 0.5 mg/dL, and the coagulation profile was normal. Urine microscopy showed numerous red blood cells with occasional atypical cells reported on cytology; there was no significant proteinuria and no casts. Serum lactate dehydrogenase was mildly elevated.

Pelvic ultrasonography of the urinary bladder. (A) A large, solid, polypoid intraluminal mass arising from the bladder base and trigone and projecting into the lumen, with a lobulated "bunch-of-grapes" surface. (B) Colour Doppler interrogation demonstrating internal vascularity within the mass. Mild bilateral pelvicalyceal fullness was noted, suggesting early lower ureteric involvement.

Pelvic ultrasonography revealed a heterogeneously echogenic, solid, grape-like polypoid mass measuring approximately 5.6 × 4.2 cm arising from the bladder base and trigone, with internal vascularity on Doppler. Both kidneys showed mild bilateral pelvicalyceal dilatation without parenchymal thinning.

Contrast-enhanced computed tomography (CECT) of the abdomen and pelvis. (A) A heterogeneously enhancing lobulated soft-tissue mass filling much of the bladder lumen and arising from the trigone/base, with clusters of grape-like fronds. (B) The mass abutting the bladder neck with mild bilateral hydroureteronephrosis; no gross perivesical fat stranding, pelvic nodal enlargement or distant lesions were seen on the study.

CECT confirmed a heterogeneously enhancing lobulated intravesical soft-tissue mass measuring approximately 6×5 cm arising from the trigone and base and causing mild bilateral hydroureteronephrosis. The perivesical planes were preserved, and there was no radiological evidence of nodal or visceral metastasis. Staging chest imaging was reported as normal.

Cystoscopic appearance. (A, B) Translucent, oedematous, grape-like polypoid fronds ("botryoid" clusters) arising from the trigone and bladder base and filling the lower bladder lumen, friable and bleeding readily on contact.

Cystoscopy demonstrated multiple soft, translucent, grape-like polypoid clusters arising from the trigone and bladder base, friable and haemorrhagic on contact, characteristic of a botryoid lesion rather than a papillary urothelial tumour.

Operative Procedure

After transfusion and stabilisation, the child underwent examination under anaesthesia and transurethral biopsy of the polypoid fronds to establish a tissue diagnosis, with control of bleeding points by careful coagulation. Given the trigonal and bladder-neck involvement, definitive resection was deliberately deferred pending histopathology and multidisciplinary planning, in keeping with a bladder-preservation, chemotherapy-first strategy. Adequate deep and superficial biopsy fragments were submitted for histopathology and immunohistochemistry.

Endoscopic and gross specimen. (A, B) Endoscopic biopsy of the botryoid fronds at the trigone. (C) Grape-like tissue fragments retrieved on resection. (D) Gross photograph of the friable polypoid specimen submitted for histopathology.

Histopathology

Microscopic examination showed a polypoid lesion covered by attenuated urothelium beneath which lay a densely cellular, condensed "cambium layer" of small round-to-spindle tumour cells overlying a hypocellular myxoid stroma. Scattered rhabdomyoblasts with eccentric eosinophilic cytoplasm, and occasional elongated strap cells with cross-striations, were identified. On immunohistochemistry the tumour cells were positive for desmin, myogenin (MYF4) and MyoD1, confirming skeletal-muscle differentiation. The features were diagnostic of botryoid embryonal rhabdomyosarcoma of the urinary bladder.

Figure 5. Photomicrographs (haematoxylin and eosin, with immunohistochemistry). Sections show a subepithelial condensed cambium layer of primitive round and spindle cells over a myxoid stroma, with scattered rhabdomyoblasts and rare strap cells. Inset: diffuse nuclear positivity for myogenin.

Outcome and Follow-up

The child was referred to paediatric oncology and staged as a localised, group III (gross residual, biopsy only) bladder/prostate-region embryonal rhabdomyosarcoma. He was commenced on multi-agent neoadjuvant chemotherapy (vincristine, actinomycin-D and cyclophosphamide) with a bladder-preserving intent, and a plan for reassessment imaging and delayed conservative surgery with or without radiotherapy depending on response. The haematuria settled and hydronephrosis improved after the first cycles. Regular clinical, cystoscopic and imaging follow-up was arranged to monitor response, organ preservation and recurrence.

Case 3. Inflammatory Myofibroblastic Tumour of the Urinary Bladder

Case Presentation

A 9-year-old girl presented with a ten-day history of gross haematuria with clots, suprapubic discomfort and increased urinary frequency with dysuria. There was no fever, flank pain, weight loss or history of trauma, and no significant past medical, surgical or family history; there had been no recent instrumentation of the urinary tract. On examination she was afebrile and haemodynamically stable, with mild suprapubic tenderness and no palpable mass. The skin and systemic examination were unremarkable.

Investigations

Laboratory evaluation revealed moderate anaemia with a haemoglobin of 8.6 g/dL and a haematocrit of 26.8%. The total leucocyte count was $13,400/\text{mm}^3$ and the platelet count $4.6 \text{ lakh}/\text{mm}^3$ ($460 \times 10^9/\text{L}$), consistent with a reactive thrombocytosis. Blood urea was 24 mg/dL and serum creatinine 0.5 mg/dL, and the coagulation profile was normal. Inflammatory markers were raised, with an erythrocyte sedimentation rate of 58 mm/hr and elevated C-reactive protein. Urine microscopy showed numerous red blood cells and a few leucocytes with mild proteinuria; urine culture was sterile and cytology was negative for malignant cells.

Pelvic ultrasonography of the urinary bladder. (A) A broad-based, hypoechoic, solid mural mass arising from the right lateral bladder wall and projecting into the lumen, measuring approximately 4.1×3.4 cm. (B) Colour Doppler showing moderate internal vascularity. Both kidneys were normal without hydronephrosis or calculi. Ultrasonography revealed a broad-based solid mural mass measuring approximately 4.1×3.4 cm arising from the right lateral bladder wall, with moderate internal vascularity on Doppler. Both kidneys were grossly normal, with no hydronephrosis or calculi.

Contrast-enhanced computed tomography (CECT) of the kidneys, ureters and bladder. (A) A well-defined, heterogeneously and avidly enhancing polypoid mural mass arising from the right lateral/posterolateral bladder wall measuring approximately 4.5×3.5 cm, with focal wall thickening at its base raising suspicion of a neoplastic growth. (B) Preserved perivesical fat planes with no pelvic lymphadenopathy or distant lesions. Both kidneys were normal. CECT confirmed a heterogeneously enhancing polypoid mural mass measuring approximately 4.5×3.5 cm arising from the right posterolateral bladder wall, with focal thickening at its base and preserved perivesical planes. There was no pelvic nodal enlargement, and both kidneys were normal. The enhancing solid mass was regarded as suspicious for a neoplastic lesion, and malignancy could not be excluded on imaging.

Cystoscopic appearance. (A, B) A solitary, fleshy, broad-based polypoid mass with a smooth-to-nodular surface and focal surface ulceration arising from the right lateral wall, bleeding on contact. Cystoscopy revealed a solitary fleshy, broad-based polypoid mass arising from the right lateral wall with focal surface ulceration that bled readily on contact. The appearance was that of a solid mural neoplasm rather than a vascular submucosal lesion, and biopsy was performed.

Operative Procedure

After correction of anaemia, and following a biopsy that raised a spindle-cell lesion in the differential, the patient underwent open surgical exploration through a lower midline abdominal incision, with the bladder approached by an extraperitoneal route. A solitary firm mural mass was confirmed at the right posterolateral wall. A partial cystectomy was performed, excising the lesion with a rim of macroscopically normal bladder wall, taking care to preserve the ipsilateral ureteric orifice; the resection margins were confirmed clear on frozen section. The bladder was closed in layers over a urethral catheter and the specimen sent for histopathology.

Intraoperative photographs. (A, B) Open extraperitoneal exposure of the bladder with the firm mural mass at the right posterolateral wall. (C) Partial cystectomy with a rim of normal bladder wall after protecting the ureteric orifice. (D) Gross excised specimen showing a solid, whorled, greyish-white cut surface.

Histopathology

Microscopic examination showed a proliferation of bland spindle-shaped myofibroblastic cells arranged in loose fascicles within a myxoid and oedematous stroma, admixed with a prominent inflammatory infiltrate of plasma cells, lymphocytes and scattered eosinophils, together with delicate thin-walled vessels. Mitotic figures were sparse and there was no atypical mitosis, coagulative necrosis or nuclear pleomorphism to suggest malignancy. On immunohistochemistry the spindle cells were positive for smooth-muscle actin and vimentin, showed cytoplasmic positivity for anaplastic lymphoma kinase (ALK), and were negative for pan-cytokeratin, desmin (predominantly) and myogenin. The features were diagnostic of an inflammatory myofibroblastic tumour of the urinary bladder.

Photomicrographs (haematoxylin and eosin, with immunohistochemistry). Sections show fascicles of bland myofibroblastic spindle cells in a myxoid stroma with a dense plasma-cell and lymphocytic infiltrate and scattered eosinophils. Inset: cytoplasmic ALK positivity in the spindle-cell component.

Outcome and Follow-up

The postoperative course was uneventful, the catheter was removed after a satisfactory cystogram, and the haematuria resolved completely. The inflammatory markers and thrombocytosis normalised over the following weeks. Because inflammatory myofibroblastic tumour is a neoplasm of intermediate biological potential with a recognised, if low, tendency to local recurrence, the child was enrolled in regular clinical, ultrasonographic and cystoscopic surveillance to detect recurrence or residual disease. No adjuvant therapy was required following complete excision.

DISCUSSION

Haemangioma of the urinary bladder is a rare benign tumour that constitutes roughly 0.6% of all bladder tumours and is most often congenital, arising from embryonic angioblastic stem cells.^{1,2} It may occur at any age but is uncommon in children and demonstrates a slight male predominance. In the published literature

most lesions are solitary (about 66%) and of the cavernous type, ranging from a few millimetres to 10 cm and favouring the dome, posterior wall and trigone.² In

contrast, the present case was notable for multiple lesions ranging from 2 to 5 cm confined to the dome.

The predominant clinical symptom is recurrent, isolated gross haematuria; suprapubic pain from vesical irritation and urinary retention are also described, and severe bleeding may cause anaemia or haemorrhagic shock.³ Our patient presented with lower abdominal pain and gross haematuria and, importantly, a comparable self-limiting episode seven years earlier — a pattern of recurrence typical of untreated bladder haemangioma.

Three histological subtypes are recognised: the cavernous form is the most common, followed by the capillary and arteriovenous subtypes. Histologically the tumour is composed of confluent, blood-engorged, dilated vessels lined by a flattened endothelium, with vessel walls occasionally thickened by adventitial fibrosis² — findings that corresponded closely with those in our specimen.

Bladder haemangioma may coexist with cutaneous haemangiomas and with the Klippel-Trenaunay-Weber and Sturge-Weber syndromes, so systemic evaluation of affected patients is strongly recommended.⁴ In the index case no such associated findings were identified. The differential diagnosis of a vascular-appearing bladder mass is broad and includes other vascular tumours, inflammatory pseudotumour, leiomyoma, phaeochromocytoma, bladder papilloma, transitional cell carcinoma, rhabdomyosarcoma and neurofibromatosis; the radiological suspicion of malignancy in our patient underscores this diagnostic difficulty.

A range of imaging modalities may contribute to diagnosis — ultrasonography with Doppler, intravenous pyelography, computed tomography, magnetic resonance imaging and pelvic angiography — but cystoscopy is central to identifying the characteristic bluish-red submucosal lesion, and an accurate diagnosis ultimately requires histological confirmation by biopsy. Management is controversial and should be tailored to lesion size and depth of penetration because of the bleeding risk of these highly vascular lesions. Options range from observation to transurethral resection, electrocoagulation, laser therapy and partial or complete cystectomy. Asymptomatic haemangiomas do not require treatment, but follow-up remains important to detect recurrence or residual disease. Severe abdominal pain, haematuria causing anaemia and suspicion of malignancy are accepted indications for surgery. In our patient, progressive anaemia together with a large vesical mass prompted operative removal with a safe margin, after which the haematuria resolved.

CONCLUSION

Bladder haemangioma is a rare tumour, and information about its characteristics remains limited. Its clinical presentation has no pathognomonic signs, although gross haematuria is the most frequent complaint. Management is controversial owing to the bleeding risk of this highly vascular lesion; small lesions may be treated by transurethral resection or laser fulguration, whereas

larger, symptomatic or multiple lesions — as in our patient — may warrant open excision with a safe margin. Despite its benign nature and slow growth, recurrent haematuria persists if the lesion remains untreated. Bladder haemangioma should therefore be included in the differential diagnosis of recurrent gross haematuria in children, with cystoscopy and histopathology forming the cornerstone of diagnosis.

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