Urinary bladder cavernous hemangioma in a 3-year-old: A rare case report

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Abstract
Cavernous hemangioma (CH) of urinary bladder occurs relatively infrequently, accounting for 0.6% of all bladder tumors. This tumor may occur sporadically or coexist with other benign and malignant vascular lesions. In this report, we present a rare case of CH in a 3-year-old Ugandan girl. A 3-year-old girl was referred to Mbarara Regional Referral Hospital (MRRH) for urological evaluation following a 3-year history of intravaginal swelling, dysuria, and heavy hematuria resulting in anemia. Imaging was consistent with polypoid bladder mass arising from the bladder trigone. Embryonal rhabdomyosarcoma was suspected based on clinical eyeballing. She was worked up for chemotherapy and received 26 cycles of vincristine sulfate, actinomycin-d, and cyclophosphamide (VAC). Biopsy and fulguration were performed after optimizing the patient. Histopathology confirmed CH. The surgery was uneventful and resulted in complete cure. CH should be considered in the differential diagnosis of childhood genitourinary masses. It is a rare entity in the real-life clinical practice and therefore can be overlooked. Excision biopsy and histology should be performed before initiating the patients to chemotherapy. CH is very insensitive to chemotherapy and therefore surgery maybe adequate in resource-limited settings.

Keywords
cavernous hemangioma, embryonal rhabdomyosarcoma

1 | INTRODUCTION

Cavernous hemangioma of urinary bladder occurs relatively infrequently, accounting for 0.6% of all bladder tumors with few cases reported in the literature (Table 1). Bladder hemangiomas result from embryological remains of unipotent angioblastic cells (of mesenchymal origin) that develop in an anomalous way inside the blood vessels. Most urinary bladder CH are solitary and smaller than 3 cm in diameter, affecting the dome, posterior wall, or trigone of the bladder. This tumor may occur sporadically or coexist with other benign and malignant vascular lesions such as cutaneous hemangiomas, Klippel–Trenaunay syndrome, Sturge–Weber syndrome, encephalo-trigeminal-angiomatosis, Osler–Weber–Rendu disease, hemorrhagic telangiectasia syndrome...
and systemic angiomatosis.\textsuperscript{1,2,4,7} The tumor presents as an incidental finding during workup for hematuria and/or voiding symptoms.\textsuperscript{8} It can be differentiated from malignancy by its lack of cytologic atypia and mitotic cytonuclear.\textsuperscript{9–11} CH should be considered in the differential diagnosis of childhood genitourinary masses.\textsuperscript{9,11} Herein, we describe a case of urinary bladder CH in a 3-year-old female Ugandan.

2  |  CASE PRESENTATION

2.1  |  Presenting symptoms

A 3-year-old girl was referred to Mbarara Regional Referral Hospital (MRRH) for urological evaluation following a 3-year history of intravaginal swelling, dysuria, and heavy hematuria resulting in recurrent anemia. There

| Table 1 | Clinical and radiological findings of carvenous hemangioma included in the review of the literature |
|---------|------------------------------------------------------------------------------------------------|
| Author  | Type of study | Number of cases | Symptoms                  | Site of hemangiomas                          | Surgical intervention                                                                 | Country   |
|---------|---------------|-----------------|---------------------------|---------------------------------------------|----------------------------------------------------------------------------------------|-----------|
| Syu et al., 2019\textsuperscript{1}  | Case report  | 1               | Hematuria                 | Anterior wall of the urinary bladder        | En bloc resection of the urachus and bladder tumor                                     | Taiwan    |
| Zhao et al., 2020\textsuperscript{2} | Case reports | 2               | Hematuria                 | First case: Multiple nodules and masses      | First case: Radical cystectomy was performed with open-methods surgery. Second case: Laparoscopic partial cystectomy | China     |
| Kim et al., 2015\textsuperscript{3}   | Case reports | 1               | Hematuria                 | Polypoid Intraluminal                       | Cold-cup biopsy, coagulated with a Holmium laser                                        | Korea     |
| Jibhkate et al., 2015\textsuperscript{4} | Case report | 1               | Hematuria                 | Dome                                        | Cystectomy and open cystectomy                                                       | India     |
| Correa-Rivas et al., 2011\textsuperscript{5} | Case report | 1               | Hematuria                 | Ovarian Torsion hematuria                   | Right salpingo-ophorectomy                                                             | USA       |
| Castro, V., F.B. Sarria, and R. Trujillo, 2011\textsuperscript{6} | Case report | 1               | Hematuria                 | –                                            | Cystoscopy + resection                                                                 | Spain     |
| Sajitha et al., 2020\textsuperscript{7} | Case report | 1               | Hematuria                 | Ureter                                      | Radical nephrectomy                                                                   | India     |
| Rastogi et al., 2010\textsuperscript{8} | Case report | 1               | Hematuria                 | Vesicoureteric junction                     | A transurethral resection with cystoscopy                                             | India     |
| Ikeda et al., 2004\textsuperscript{9}  | Case report  | 1               | Hematuria                 | Bladder wall-non specific                   | Cystoscopy and transurethral resection                                                | Japan     |
| Tornero Ruiz, J.I., et al., 2006\textsuperscript{10} | Case report | 1               | Hematuria                 | Rectovesical                                | Cystoscopy                                                                         | Spain     |
| Sousa et al., 2017\textsuperscript{11} | Case report  | 1               | Hematuria                 | Bladder Dome                                | Radical cystectomy                                                                  | Brazil    |
| Numanoğlu and Tatlı, 2008\textsuperscript{12} | Case report | 1               | Shock                     | Bladder Dome                                | Cystostomy and open cystectomy                                                       | Turkey    |
| KIM, G.-P., et al., 1999\textsuperscript{13} | Case report | 1               | Hematuria                 | –                                           | Partial cystectomy                                                                   | Korea     |
| Cheng et al., 1999\textsuperscript{14}  | Case series  | 19              | Hematuria                 | Bladder Dome                                | Cystoscopy and transurethral resection-14 Biopsy and fulguation-5                    | India     |
| Msakni et al., 2016\textsuperscript{15} | Case report  | 1               | Hematuria                 | Right lateral wall of the bladder           | A transurethral resection with cystoscopy                                              | Tunisia    |
| Tavora et al., 2008\textsuperscript{16} | Case series | 13              | Hematuria and fever        | –                                           | A transurethral resection with cystoscopy                                              | USA       |
was a history of recurrent blood transfusion, six times before referral to our facility.

2.2 | Past medical history

Occasionally treated for febrile illness that the mother attributed to malaria though there were no medical reports to confirm. Swelling protruding through the vagina compelled the mother to visit traditional healthcare provider before reporting to several health facilities.

2.3 | Physical examinations

Severe pallor of the conjunctivae, febrile with a temperature of 39.6°C. There was no scleral icterus. Local examinations revealed pendulous mass, arising from the urethral meatus held with a stalk and easily bleeds on touching (Figure 1A). Per abdomen: Mild to moderate suprapubic distention and tenderness, dull on percussion.

3 | INVESTIGATIONS

3.1 | Blood workup and urinalysis

There were neutrophilic leukocytosis (of $34.48 \times 10^3/\mu l$), thrombocytosis (of $375 \times 10^3/\mu l$), and anemia of 4.6 g/dl. Significantly in urinalysis were hemoglobinuria, proteinuria, and leukocytes of more than 10 white blood cells/high field (Table 2).

3.2 | Ultrasound scan

Pelvic ultrasound scan showed mildly thickened bladder walls with mobile heterogeneously echogenic solid mass seen in the urinary bladder lumen measuring $(5.17 \times 4.10 \times 4.4 \text{ cm})$ with a volume of $49.2\text{cc}$ (Figure 2A). The left kidney demonstrated moderately dilated fluid-filled calyces, renal pelvis, and proximal ureters (Figure 2B). No focal masses or nephrolithiasis were seen. Right kidney $(7.15 \times 2.83 \text{ cm})$ and left kidney $(8.03 \times 3.81 \text{ cm})$. The uterus and adnexae were normal, free of masses, and fluid collections.

3.3 | Computed tomography (CT)

CT showed a large lobulated polypoid intravesical heterogeneously enhancing mass, appeared to be arising from urinary bladder trigone measuring $5.22 \times 4.80 \times 7.13 \text{ cm}$ in its widest dimensions (Figure 3A,B). Axial and coronal CECT images showed grossly dilated left ureter, pelvis, and superior calyx (Figure 4A,B). There was obliteration of space of Retzius, vesicouterine, rectouterine, vesicovaginal, and rectovaginal spaces due to mass effect.

4 | TREATMENTS

4.1 | Medication and chemotherapy

Antibiotics (metronidazole and ceftriaxone) were initiated for two weeks. The child also received three units of whole blood before initiating on vincristine, dactinomycin, and cyclophosphamide (VAC) which she received 26 cycles.

4.2 | Surgery and postoperative treatment

Examination under anesthesia (Figure 1B), excision, and fulguration of the mass was performed under general anesthesia. The child was given general anesthesia and positioned in a lithotomy position and in aseptic technique draped. Urethral meatus was visualized after retraction and catheterized with Foley’s catheter size 8 Fr (drained bloody urine). We found a foul-smelling polypoid mass protruding from the urethra, highly friable with areas of necrosis (Figure 1B). Excision of the mass

**Figure 1** (A) Polypoid mass (PM), Vagina (V), and urethra (U) viewed during physical examination. (B) Followed examination under anesthesia (EUA), excision and fulguration was performed
at the stalk was performed with monopolar diathermy taking care and sparing the urethra. The patient received one more unit of whole blood intraoperative. The patient continued ceftriaxone and intravenous paracetamol for the next one week. Postoperative period was uneventful. She was discharged after one week following surgery on oral antibiotics and hematinic as the histology report was to follow.

### TABLE 2 Bloodwork up and urinalysis

| Parameters                  | Laboratory values at admission | Laboratory values in the last follow-up | Reference point |
|-----------------------------|-------------------------------|----------------------------------------|-----------------|
| Hemoglobin g/dl             | 4.6                           | 11.2                                   | (9.5–13.5)      |
| Hematocrit (%)              | 24.2                          | 36                                     | (35–44)         |
| White cell counts [10⁹/µl]  | 34.48 x 10³                   | 8.4 x 10³                              | (5.50–17.00)    |
| Neutrophils                 | 18.2 x 10⁹                    | 4.6 x 10⁹                              | (1.50–7.00)     |
| Platelets                   | 375 x 10³                     | 340 x 10³                              | (150–400)       |
| Creatinine (mg/dl)          | 0.9                           | Not repeated                           | (0.6–1.1) Females (0.7–1.3) Males |
| Alanine transaminase (u/L)  | 12                            | Not repeated                           | (0–42)          |
| Aspartate transaminase (u/L)| 17                            | Not repeated                           | (0–37)          |
| Sodium (mmol/L)             | 129                           | Not repeated                           | (135–145) mmol/L |
| Potassium (mmol/L)          | 4.1                           | Not repeated                           | (3.5–5.5) mmol/L |
| Calcium(mmol/L)             | 107                           | Not repeated                           | (1.1–3.35) mmol/L |
| Chloride (mmol/L)           | 91                            | Not repeated                           | (95–105) mmol/L |
| Urinalysis                  |                               |                                        |                 |
| Red blood cells             | >100 red blood cells/high power field | Negative                             |                 |
| Leukocytes                  | >10 white blood cells/ high power field | Negative                             |                 |
| Protein                     | Proteinuria                   | Negative                               |                 |

**FIGURE 2** Transverse and longitudinal abdominal ultrasound images show lobulated heterogeneous solid mass in the urinary bladder lumen (A) and moderately dilated left renal calyces (B)

### 4.3 Histopathology

Received a single piece of firm dark brown with light brown areas (1.2 x 0.8 x 0.3) cm (**Figure 5A,B**). Sections show extravascular hemorrhage, numerous dilated vascular channels, some of which had thrombosis, and a thin wall. No immature rhabdoblasts consistent with vascular lesion (CH).
4.4 | Follow-up

The patient was referred to a national hospital, but the mother declined because of financial constraints. Follow-up ultrasound images demonstrated a normal urinary bladder with no residual tumor (Figure 6A) and normal kidneys with resolution of previously demonstrated left moderate hydroureteronephrosis (Figure 6A,B). Repeated urinalysis was normal.

5 | DISCUSSION

Hemangioma of the urinary bladder is the second most common hemangioma of the urinary tract system, a case series of 19 patients is the largest published in the literature to date. Most are solitary and of the cavernous type, occurring on the posterior wall or dome of the urinary bladder. In the present case, the patient presented with dysuria, hematuria, and recurrent anemia which is consistent with case in published literature. Isolated hematuria results from thrombus, infarction, and angiogenesis by the erosion of the urothelium. Other symptoms include suprapubic pain due to vesicle irritation and urinary retention which may eventually result in obstructive uropathy which was a case for this patient. This is similar to other study. Our case showed a mobile heterogeneously echogenic solid mass, with mildly thickened urinary bladder wall at 0.45cm. Hydroureteronephrosis can occur as a result of ureteric obstruction by the mass, and a hematom
can obscure the mass in the bladder when there is massive bleeding. Mild left hydroureteronephrosis was also seen in this case due to mass effect. Computed tomography (CT), pelvic arteriography, and magnetic resonance imaging are useful in defining and location of a hemangioma but are not helpful diagnostic tools in distinguishing bladder hemangiomas from other bladder tumors.

In our setting, however, we can only perform CT and ultrasonography for any pelvic mass. Cystoscopy usually reveals a reddish-blue, polypoid, submucosal mass with intact urothelium; however, we were not able to perform cystoscopy because of lack of its availability.

Treatment of CH depends on, size, location, and depth of penetration. For small lesions and asymptomatic hemangiomas, surveillance is sufficient. The treatment is only necessary when the lesions threaten the organ function or the patients' performance status, such as hematuria, anemia, and suspicion of malignant lesion. Our case developed recurrent anemia, obstructive uropathy and urosepsis (evidence by neutrophilic leukocytosis), justifying surgical approach. Treatment options usually involve observation, transurethral resection, electrocoagulation, radiation, systemic steroids administration, injection of a sclerosing agent, interferon-alpha-2 therapy, laser therapy, and partial cystectomy or complete cystectomy. Bladder CH is histologically similar to hemangiomas found at other sites with numerous proliferative capillaries mixed with thin-walled, dilated blood-filled vessels lined with flattened endothelium. The vessels are sometimes thickened by adventitial fibrosis.

6 | CONCLUSION

Patients with CH of the urinary bladder have a favorable outcome. CH should be considered in the differential diagnosis of childhood genital embryonal rhabdomyosarcoma. Excision biopsy and histology should be performed.
before initiating the patients to chemotherapy to avoid toxicities. Although CH are rare benign tumors, it should be kept in mind that they can lead to life-threatening complications when the diagnosis is delayed. CH is insensitive to chemotherapy and, therefore, surgery is effective when the lesion is small.

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CONFLICT OF INTEREST
No conflict of interest.

AUTHOR’S CONTRIBUTIONS
O.C.N performed conceptualization, data acquisition, drafting of the case report and final approval of the version to be published. AR evaluated the pathology of this case. P.A.O, E.O, and M.A interpreted radiological imaging. B.F was the main editor of the final case report. F.K edited the first draft of the work, and M. S supervised surgery during excision and biopsy and approved the final version to be published.

ETHICAL APPROVAL
Ethical clearance was obtained from Mbarara University Faculty of Medicine Research Committee, MUST-29/06-21.

INFORMED CONSENT
The mother provided informed consent for photographs, procedures, and tissue processing.

CONSENT FOR PUBLICATION
The mother provided an informed written consent for this case to be published in a peer-reviewed journal.

DATA AVAILABILITY STATEMENT
All underlying data have been supplied in the manuscript.

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