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CASE REPORT

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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[1–3] 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.[4,5] Most urinary bladder CH are solitary and smaller than 3 cm in diameter, affecting the dome, posterior wall, or trigone of the bladder.[5,6] 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.[1,2,4,7] The tumor presents as an incidental finding during workup for hematuria and/or voiding symptoms.[8] It can be differentiated from malignancy by its lack of cytologic atypia and mitotic cytonuclear.[9–11] CH should be considered in the differential diagnosis of childhood genitourinary masses.[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[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[2]	Case reports	2	Hematuria	First case: Multiple nodules and masses Second case: Right anterior and superior wall of the bladder	First case: Radical cystectomy was performed with open-methods surgery. Second case: Laparoscopic partial cystectomy	China
Kim et al., 2015[3]	Case reports	1	Hematuria	Polypoid Intraluminal	Cold-cup biopsy, coagulated with a Holmium laser	Korea
Jibhkate et al., 2015[4]	Case report	1	Hematuria	Dome	Cystectomy and open cystectomy	India
Correa-Rivas et al., 2011[5]	Case report	1	Ovarian Torsion hematuria	Ovary	Right salphingo-ophorectomy	USA
Castro, V., F.B. Sarria, and R. Trujillo, 2014[6]	Case report	1	Hematuria	–	Cystoscopy + resection	Spain
Sajitha et al., 2020[7]	Case report	1	Hematuria	Ureter	Radical nephrectomy	India
Rastogi et al., 2010[8]	Case report	1	Hematuria	Vesicoureteric junction	A transurethral resection with cystoscopy	India
Ikeda et al., 2004[9]	Case report	1	Hematuria	Bladder wall-non specific	Cystoscopy and transurethral resection	Japan
Tornero Ruiz, J.I., et al., 2006[10]	Case report	1	Hematuria	Rectovesical	Cystoscopy	Spain
Sousa et al., 2017[11]	Case report	1	Hematuria	Bladder Dome	Radical cystectomy	Brazil
Numanoğlu and Tatlı, 2008[12]	Case report	1	Shock	Bladder Dome	Cystostomy and open cystectomy	Turkey
KIM, G.-P., et al.,1999[13]	Case report	1	Hematuria	–	Partial cystectomy	Korea
Cheng et al., 1999[14]	Case series	19	Hematuria	Bladder Dome	Cystoscopy and transurethral resection-14 Biopsy and fulguation-5	India
Msakni et al., 2016[15]	Case report	1	Hematuria	Right lateral wall of the bladder	A transurethral resection with cystoscopy	Tunisia
Tavora et al., 2008[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 ($5.17 \times 4.10 \times 4.4$ cm) with a volume of 49.2cc (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 (7.15×2.83) cm and left kidney (8.03×3.81) 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 rising from urinary bladder trigone measuring $5.22 \times 4.80 \times 7.13$ 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

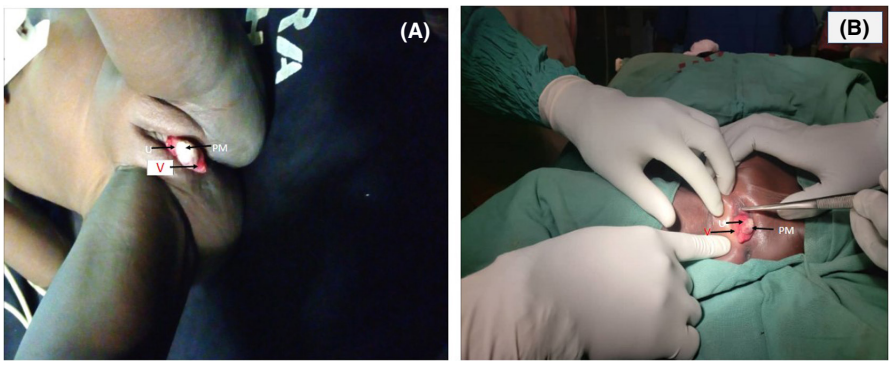

FIGURE 1 (A) Polypoid mass (PM), Vagina (V), and urethra (U) viewed during physical examination. (B) Followed examination under anesthesia (EUA), excision and fulguration was performed

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 [$10^3/\mu l$]	34.48×10^3	8.4×10^3	(5.50–17.00)
Neutrophils	18.2×10^3	4.6×10^3	(1.50–7.00)
Platelets	375×10^3	340×10^3	(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	

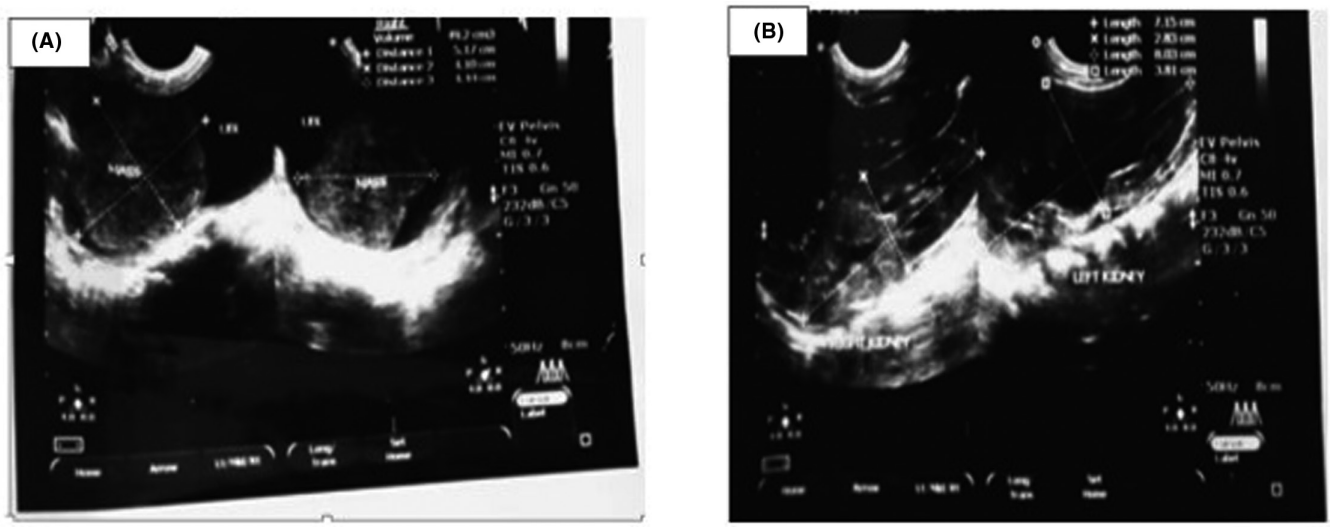

FIGURE 2 Transverse and longitudinal abdominal ultrasound images show lobulated heterogeneous solid mass in the urinary bladder lumen (A) and moderately dilated left renal calyces (B)

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.

4.3 | Histopathology

Received a single piece of firm dark brown with light brown areas (1.2 × 0.8 × 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).

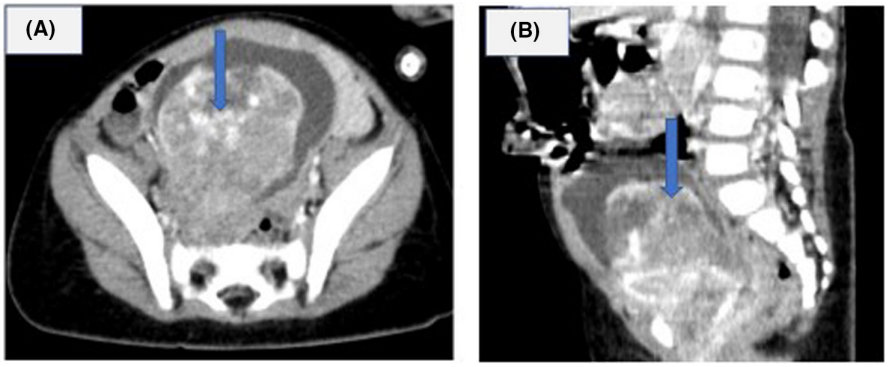

FIGURE 3 (A and B) Axial and sagittal CECT images show a large lobulated polypoid intravesical heterogeneously enhancing mass, which appears to arise from the trigone of the urinary bladder (blue arrows)

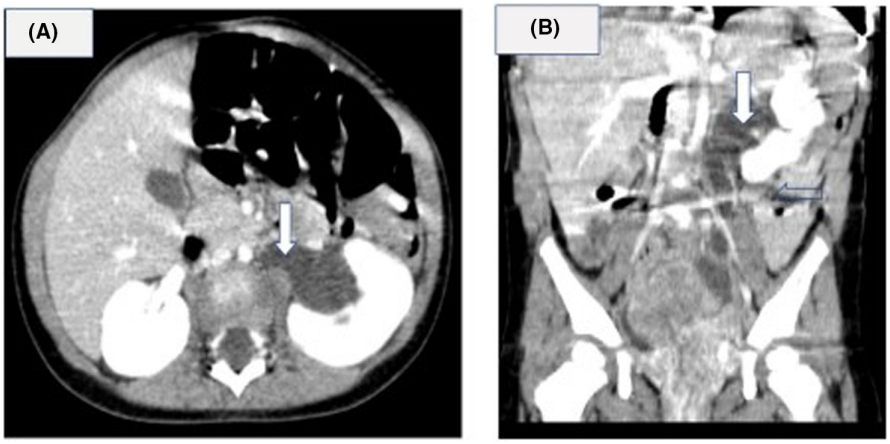

FIGURE 4 Axial (A) and coronal (B) CECT images show left hydroureteronephrosis (white arrows)

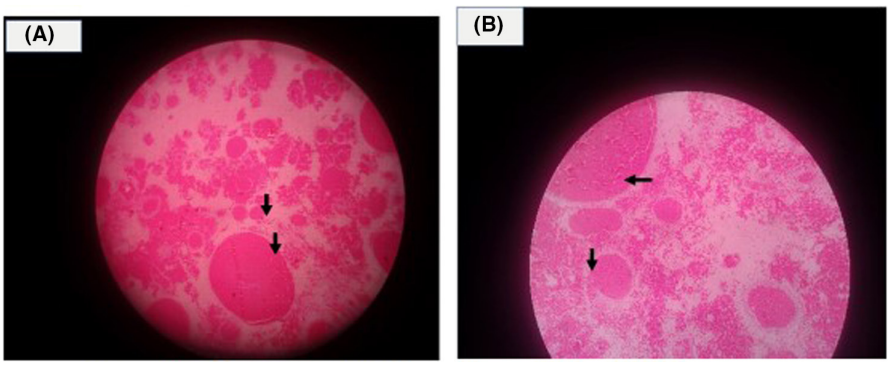

FIGURE 5 Numerous dilated vascular channels laden with red blood cells indicated in black arrows in A and B

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.[9] Most are solitary and of the cavernous type, occurring on the posterior wall or dome of the urinary bladder.[9] In the present case, the patient presented with dysuria, hematuria, and recurrent anemia which is consistent with case in published literature.[4,7] 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.[4,12] This is similar to other study.[3] 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 hematoma

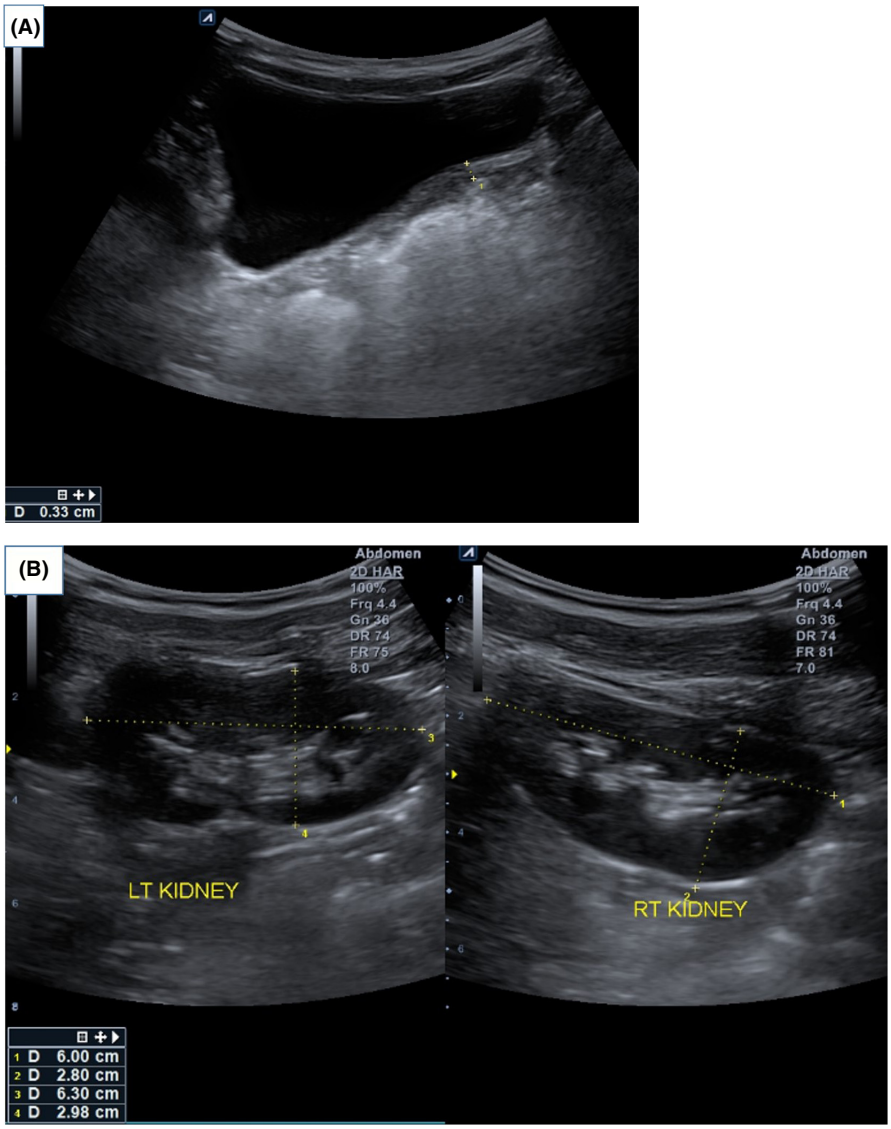

FIGURE 6 Follow up abdominopelic ultasound scan.Normal urinary bladder(6A), and resolution of hydroureteronephrosis(6B)

can obscure the mass in the bladder when there is massive bleeding.[3] 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.[2]

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[2,13]; 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.[14] 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.[2,15] 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.[16] The vessels are sometimes thickened by adventitial fibrosis.[1,7]

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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