**Case report**

**A Rare Case of Life-Threatening Severe Haematuria in a Young Female due to Angiosarcoma of the Bladder: A Case Report and Literature Review**

**Abstract**

**Background**: Angiosarcoma of the bladder is a rare and aggressive malignancy arising from malignant endothelium accounting for less than 2% of all sarcomas. It typically affects older adults, with few cases reported in younger individuals. The rarity and nonspecific clinical presentation make diagnosis and management particularly challenging.

 **Case Presentation**: We report the case of a 32-year-old female who presented with life-threatening haematuria and anaemia. Initial assessment suggested bladder clots, and diagnostic cystoscopy was inconclusive due to persistent bleeding and extensive clot formation. The patient underwent emergency laparotomy, during which a biopsy from the thickened bladder wall revealed angiosarcoma though, immunohistochemistry was not available creating a diagnostic limitation. Despite aggressive resuscitation, and internal iliac artery ligation, bleeding persisted. Once stabilised, the patient underwent an open radical cystectomy with ileal conduit diversion. Postoperative recovery was initially uneventful, but the patient later succumbed to complications from anaemia and multi-organ failure within three months of diagnosis.

**Discussion**: This case highlights the aggressive course of bladder angiosarcoma and underscores the importance of early recognition and intervention. Due to its rarity, especially in young females without known risk factors, diagnosis is often delayed. Histopathological and immunohistochemical evaluation remain crucial for definitive diagnosis. Management requires a multimodal approach, often including radical surgery, and possibly chemotherapy or radiotherapy depending on the disease stage. Despite timely intervention, prognosis remains poor with high mortality.

 **Conclusion:** Bladder angiosarcoma, although rare, should be considered in the differential diagnosis of unexplained haematuria, even in young patients. Prompt diagnosis and radical treatment are vital, although the prognosis remains guarded.

**Introduction**
Angiosarcoma of the bladder is an exceedingly rare and aggressive malignancy characterised by the proliferation of malignant endothelial cells within the bladder tissue and accounting for less than 2% of sarcomas(1). Sarcomas commonly arise in adults and usually are in visceral organs, soft tissue, or bone (2). Due to their rarity, these tumours often present diagnostic and therapeutic challenges, with limited data available regarding their clinical course, optimal management strategies, and long-term outcomes. The aetiology of bladder angiosarcoma remains poorly understood. However, known risk factors for angiosarcomas in general include prior radiation therapy, chronic inflammation, and exposure to carcinogens such as polyvinyl chloride and arsenic (2). Despite these associations, most cases occur sporadically without identifiable risk factors (3). De novo primary angiosarcoma of the bladder in the absence of radiation and other risk factors has only been reported in about 20 cases worldwide (4). In younger individuals, the diagnosis is particularly challenging due to the rarity of this malignancy and the nonspecific nature of its clinical presentation (4). The clinical presentation is generally nonspecific, but haematuria is the commonest symptoms. Others may present with irritative bladder symptoms like dysuria, frequency, pelvic pain etc (5)

Herein, we report a case of a 32-year-old female presenting with life-threatening haematuria who was ultimately diagnosed with angiosarcoma of the bladder. This case highlights the importance of considering rare entities in the differential diagnosis of haematuria, even in younger patients. It underscores the significance of histopathological and immunohistochemical evaluation in reaching an accurate diagnosis

**Case Presentation**

A 32-year-old female was referred to the Urology unit with severe anaemia on account of massive haematuria, which started about 5 days before the presentation. She received blood at the peripheral hospital before referral. Clinical examination reveals a young lady who is well-looking and not in any respiratory distress, anicteric, or afebrile. She was, however, severely pale.Cardiorespiratory examination was unremarkable.The abdomen was soft and non-tender with a palpable mass in the suprapubic region suspected to be a bladder mass or clots. The patient had a 3-way catheter in situ with irrigation ongoing, draining bloody urine. A manual bladder washout was done in the emergency room, a few clots were evacuated, and irrigation was initiated. Abdominal ultrasound revealed a mass in the bladder, suspected to be blood clots, with differentials of a bladder mass.The haemoglobin on admission was 6 g/dl.

The Patient was initially optimised by blood transfusion and prepped for a diagnostic cystoscopy in preparation for a possible transurethral resection of the bladder (TUR BT), which was not possible due to a huge clot in the bladder. The clot could not be evacuated endoscopically, due to persistent bleeding. An emergency laparotomy was done to evacuate the bladder clot and attempt to secure haemostasis. There was no clear mass seen intraoperatively, except some slight thickening of the bladder wall around the areas of haemorrhage. A preliminary biopsy was taken from the thickened bladder wall which revealed angiosarcoma of the bladder Postoperatively, the patient was still bleeding and was further transfused with whole blood and platelet concentrate. She had about 10 units of whole blood transfused and 4 platelet concentrates throughout her stay in the hospital.

 Renal function shows serum creatinine of 20 mg/dl, which is elevated. A full blood count also revealed an elevated white cell count of 13 x 10^9/L with differential neutrophilia. The Patient was treated with broad-spectrum antibiotics. A repeat ultrasound revealed a new clot in the bladder with bilateral hydronephrosis. The clot again could not be evacuated manually. So, a second laparotomy was done to evacuate the clots. The bleeding was persistent and active, yet the patient was not stable for cystectomy, so the anterior branch of the internal iliac artery was ligated to control the bleeding and ureterocutaneostomy was done to divert the urine. The Patient was monitored closely in the intensive care unit and later transferred to the general ward. She subsequently developed a surgical site infection at the laparotomy site and became delirious as a result of sepsis. The renal function improved with a creatinine level of 12mg/dl after the temporary urinary diversion; however, emergency dialysis was carried out daily for 3 days because the patient was in encephalopathy despite the slightly improved renal function. The encephalopathy resolved and patient was prepared for another laparotomy. An open radical cystectomy and urinary diversion using an ileum conduit were done successfully. The sample was sent for histology which reports revealed angiosarcoma which “shows a complex vascular tumour composed of variably sized vascular channels lined by atypical endothelial cells that extends from the submucosa to the inner third of the bladder wall of the bladder”. The patient was managed in the intensive care unit (ICU) and subsequently on the ward with antibiotics, fluid management, and anticoagulation. She recovered and the wound was healed, and she was discharged 2 weeks after the radical cystectomy. She spent about 8 weeks in the Hospital. The final histology confirmed an angiosarcoma of the bladder as the cause of the haematuria. Upon discharge, the patient was readmitted at a district hospital on two occasions on account of anaemia; she was transfused but unfortunately passed away on the second admission from complications of anaemia and multiorgan failure. This case illustrates the aggressive nature of angiosarcomas and the high mortality associated with them. Early diagnosis and radical treatment are necessary to guarantee survival.

**Literature Review and Discussion**

Angiosarcoma is characterised histologically by the proliferation of malignant endothelial cells lining anastomosing channels within the bladder tissue (1), as seen in the histology of this case, which shows a complex vascular tumour composed of variably sized vascular channels lined by atypical endothelial cells that extends from the submucosa to the inner third of the bladder wall. It is aggressive, demonstrated by its propensity for local invasion and metastasis (6). Most commonly, sarcomas arise in adults and usually are in visceral organs, soft tissue, or bone (3). Primary bladder angiosarcoma constitutes a particularly rare entity with most cases reported in older adults often in the sixth to eighth decades of life, and a male predominance (3,7). In this current case, the patient is a young female with bladder involvement.

Known risk factors for angiosarcomas in general include prior radiation therapy, chronic inflammation, and exposure to carcinogens such as polyvinyl chloride and arsenic (8). Our patient had no known history of exposure to any of the known risk factors, thereby placing her under the sporadic or de novo primary category (4,9). In younger individuals, the diagnosis is particularly challenging due to the rarity of this malignancy and the nonspecific nature of its clinical presentation. The patient presented with anaemia due to gross haematuria, which is the commonest presentation of angiosarcoma of the bladder (3,10). In a recent systematic review of 68 patients, 52 (76.4%) presented with haematuria as the main symptom (7). Other symptoms presented by the patient include dysuria and pelvic pain, which have also been documented as accompanying symptoms in addition to the haematuria (4). These presentations make the diagnosis very difficult because they overlap significantly with other, more common bladder pathologies, including infections, stones, and urothelial carcinoma, contributing to diagnostic delays. Imaging studies such as ultrasound, CT, or MRI may reveal a bladder mass, but are insufficient for definitive diagnosis (4). In this patient, the only radiological investigation that was readily available was an ultrasound, considering the unstable nature of the patient. A pelvic ultrasound revealed a bladder mass, suspected to be clots. The bladder wall was biopsied for histopathological examination, which remains the cornerstone for diagnosis, usually with the help of immunohistochemical staining (e.g., CD31, CD34, and Factor VIII-related antigen), which are essential for confirming the diagnosis (3). The histological findings, as stated above, could also point towards other differentials like Kaposi sarcoma, hemangioma, hemangioendothelioma, as well as atypical fibroxanthoma (11). This ultimately will make the diagnosis difficult in the absence of appropriate immunohistochemical markers. In our case, we could not do the immunohistochemical markers (CD34, CD31 and factor VIII) due to nonavailability. This brings to light the difficulties usually encountered in diagnosing this tumour and others alike in resource-limited settings like Northern Ghana.

The derangement in the renal function could be due to the hydronephrosis caused by the clots, which was demonstrated by the ultrasonography. This was more evident by the improvement in the renal function after the temporary urinary diversion (Ureterocutaneostomy). The deranged renal function, together with the sepsis from the surgical site infection, could account for the delirium or encephalopathy.

Due to the rarity of bladder angiosarcoma, there is no standardised treatment protocol (12). Management typically involves a multimodal approach, including surgery, chemotherapy, and/or radiotherapy, tailored to the patient's clinical condition and tumour stage (8). Radical cystectomy with ileal conduit is often pursued for localised disease, as was done in this case, while chemotherapy and radiation are employed in advanced or metastatic cases (8). The prognosis of angiosarcoma of the bladder is dismal, with a mean overall survival of 10.6 months, with a range of 3 days to 6 years (5). Silwal etal in their systematic review revealed that 70.4 % of 54 patient followed have a median survival of 5 months (7). Our patient died within 3 months after diagnosis from a suspected cardiac failure because of severe anaemia which is in line with literature findings. There was no autopsy done to confirm the cause of death, as this is a very common practice in rural Ghana due to inadequate number of pathologists.

**Conclusion**

In this context, our case adds valuable insights into the presentation, diagnosis, and management of bladder angiosarcoma in a young female, a demographic in which this malignancy is exceptionally uncommon. Furthermore, this report emphasises the critical role of histopathological evaluation in achieving an accurate diagnosis and guiding management decisions. Bladder angiosarcoma, although rare, should be considered in the differential diagnosis of unexplained haematuria, even in young patients.

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