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Inflammatory Myofibroblastic Tumor of the Bladder: A Case Report and Review of Literature

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ABSTRACT

Inflammatory Myofibroblastic Tumor (IMT) of the bladder is a rare benign neoplasm with fewer than 100 cases reported globally and none from Nigeria or West Africa. It is uncommon in the genitourinary tract and often mimics malignancies, making diagnosis difficult. Typically presenting as well-circumscribed, variably sized masses that can occur in various anatomical sites, though they are often found in the lungs. It can also be found in head and neck soft tissue, abdominal cavity, omentum, retroperitoneum and other tissues and organs but rarely in the genitourinary tract.

Inflammatory myofibroblastic tumor is a rare benign tumor consisting of myofibroblastic spindle cells and inflammatory cells. Immunohistochemistry, particularly anaplastic lymphoma kinase positivity, is key to distinguishing it from malignancies. Surgical excision is the primary treatment, with a generally good prognosis, although recurrence can occur in 10% -25% of cases, requiring ongoing follow-up.

We present a case of a 53-year-old diabetic Nigerian businessman, who presented with a 5-day history of gross hematuria and weakness. Despite clot evacuation, bladder irrigation, receiving blood transfusions and antifibrinolytics, his symptoms persisted. A CT (Computed Tomography) scan revealed an enhancing mass on the anterior bladder wall with no signs of metastasis. Bladder cancer was suspected and cystoscopy showed a necrotic mass and he underwent cystoscopy-guided partial cystectomy in the same setting. Post-operatively, he had prolonged wound care for surgical site infection. He has been hematuria free. Histology and immunohistochemistry confirmed IMT. He is being followed up and has had cystoscopies and urine cytology which were normal.

Keywords: Bladder, Spindle cells, Mesenchymal neoplasm

Abbreviations: IMT: Inflammatory Myofibroblastic Tumors; CT: Computed Tomography; ALK: Anaplastic Lymphoma Kinase; WHO: World Health Organization; α-SMA: alpha-Smooth Muscle Actin; TURBT: Transurethral Resection of the Bladder Tumor

1. Introduction

Inflammatory Myofibroblastic Tumors (IMTs) are rare mesenchymal neoplasms with intermediate biological potential, characterized by a proliferation of myofibroblasts and a prominent inflammatory infiltrate. While IMTs most commonly occurred in the lungs, they had been reported in various anatomical locations, including the head and neck, abdominal cavity, omentum, retroperitoneum and other soft tissues. However, IMTs were rarely encountered in the urinary tract, with the bladder being the most frequently affected site when they did occur. Due to their histological overlap with malignant spindle cell tumors, IMTs were often misdiagnosed, making early recognition and appropriate diagnostic evaluation crucial.

This case is unique because of the tumor's unusual location in the bladder, an exceedingly rare site for IMTs. Additionally, this report contributed to the limited literature on bladder IMTs, particularly in the African population, where no prior cases have been documented. The presentation of painless hematuria in a middle-aged man without significant risk factors highlighted the need for considering IMTs in the differential diagnosis of bladder masses. The case also reinforced the importance of histopathological and immunohistochemical analysis, particularly ALK expression, in confirming the diagnosis and guiding management decisions.

2. Case Presentation

A 53-year-old diabetic Nigerian businessman presented with a 5-day history of gross hematuria and weakness. Despite receiving blood transfusions and antifibrinolytics, his symptoms persisted. A CT scan revealed an enhancing mass on the anterior bladder wall with no signs of metastasis. Bladder cancer was suspected and cystoscopy showed a necrotic mass. The patient underwent partial cystectomy (Figure 1), followed by wound infection management. Histology and immunohistochemistry confirmed IMT (Figure 2 and 3). He is being followed up in the outpatient department and has had cystoscopy (Figure 4) and urine cytology done which were normal.

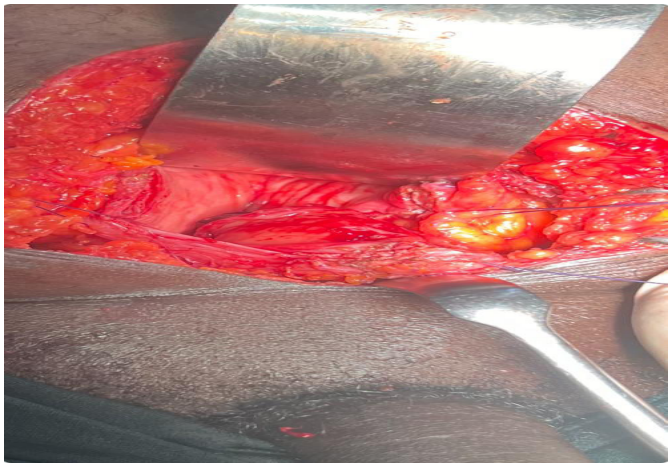

Figure 1: Intraoperative picture showing the mass in the bladder.

3. Discussion

According to World Health Organization (WHO) classification, Inflammatory Myofibroblastic Tumours (IMT) is an intermediate tumour that primarily occurs in children and young adults[1]. Inflammatory Myofibroblastic Tumors (IMTs) are mesenchymal neoplasms of intermediate biological potential. They are distinctive neoplasms characterized by a proliferation of myofibroblasts and a prominent inflammatory infiltrate. They are majorly made up of spindle cells with unclear etiology and malignant potential[2]. These tumors typically present as well-circumscribed, variably sized masses that can occur in various anatomical sites, though they are often found in the lungs. It can also be found in head and neck soft tissue, abdominal cavity, omentum, retroperitoneum and other tissues and organs. However, the IMT is rarely encountered in the urinary tract[3-5]. When seen in the urinary tract the most common site is the bladder[5].

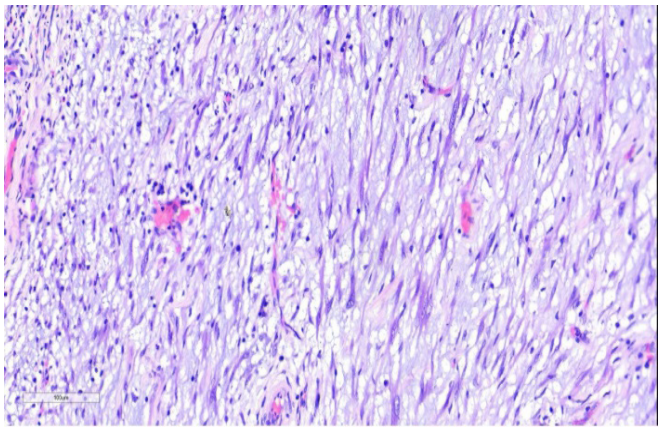

Figure 2: Micrograph showing the spindle cells within a myxoid background. Interspersed mall blood vessels and mononuclear inflammatory cells are present. X200 magnification.

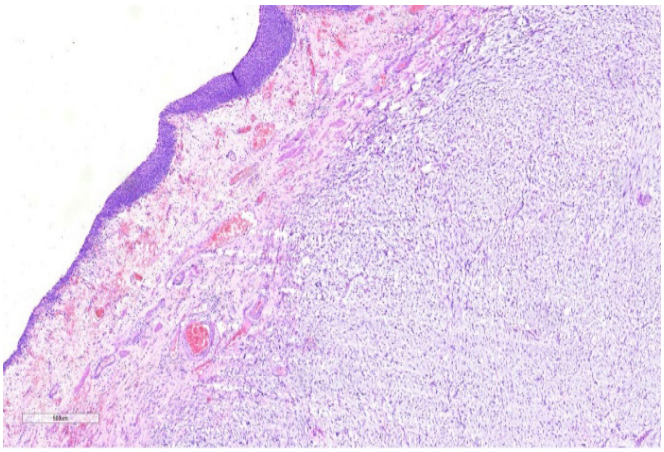

Figure 3: Micrograph showing the tumor with overlying urothelium. X40Magnifictaion.

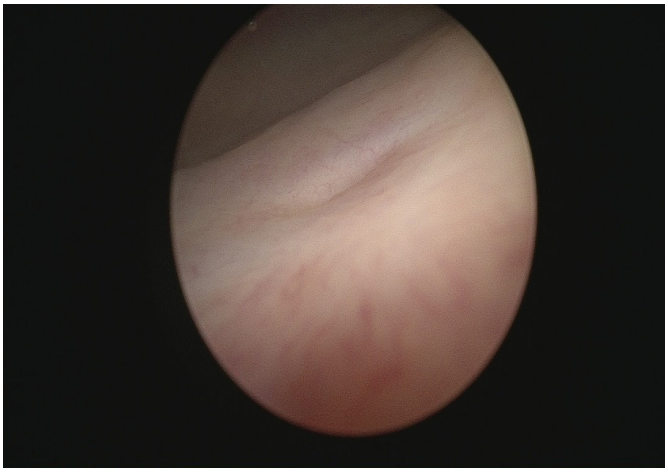

Figure 4: The 5 months post-operative cystoscopy image, no lesion seen.

They typically manifest as asymptomatic or mildly

symptomatic masses, which may be discovered incidentally during imaging for other conditions. When symptoms do occur, they can include localized pain, swelling or discomfort depending on the tumor's location. In the lungs they may present with respiratory symptoms such as cough or dyspnea. IMTs are mostly recorded in adolescents[6]. They are also often seen in children and young adults but can occur at any age[3]. The overall prevalence of IMT is 0.04% - 0.7%[7].

Histologically, IMTs are marked by a mixed inflammatory cell infiltrate, including lymphocytes, plasma cells and neutrophils, combined with a storiform pattern of myofibroblasts. Despite their aggressive histological appearance, IMTs generally exhibit a benign clinical course, though they can be locally invasive. The immunohistochemical profile of IMTs usually shows positivity for smooth muscle actin and variable expression of ALK (Anaplastic Lymphoma Kinase), which can aid in distinguishing these tumors from other similar lesions[3]. IMTs account for <1% of all bladder tumors[8]. IMTs may locally recur in 25% of patients with abdominopelvic tumors. Patients may rarely develop metastatic disease; common sites include the lung, liver, bone and brain[9].

3.1. Diagnosis

IMTs are often misdiagnosed with other malignant spindle cell tumors based on their similar cell morphology.[2] The diagnosis of Inflammatory Myofibroblastic Tumors (IMTs) begins with a clinical evaluation, patients may present with symptoms such as abdominal pain or a palpable mass, though some cases are asymptomatic and discovered incidentally during imaging[7]. Imaging often reveals well-circumscribed lesions that enhance with contrast. Definitive diagnosis requires histopathological examination through a biopsy, which shows a proliferation of myofibroblasts along with a mixed inflammatory infiltrate, typically exhibiting a fibromatosis-like pattern[4]. Immunohistochemistry reveals markers like alpha-Smooth Muscle Actin (α-SMA) and, in certain cases, the presence of ALK protein, confirming the diagnosis. However, differentiating IMTs from other mesenchymal tumors poses challenges due to overlapping features, necessitating a multidisciplinary approach for accurate assessment and effective management[10]. Grossly, Inflammatory Myofibroblastic Tumors (IMTs) typically present as white or tan masses with a firm, fleshy or gelatinous texture. Histologically, they are characterized by spindle cells interspersed with varying degrees of inflammatory cell infiltration, arranged in myxoid, compact or hyalinized patterns. The myxoid and compact patterns refer to loosely and densely packed spindle cell formations, respectively, while the hyalinized pattern features elongated spindle cells alongside sparse collagen fibers.

3.2. Treatment

The management of Inflammatory Myofibroblastic Tumors (IMTs) primarily involves surgical resection, which is considered the first-line treatment[8]. A systematic review of total of 75 case reports on bladder IMTs showed Transurethral Resection of the Bladder Tumor (TURBT) (34%), to commonly done procedure followed by complementary partial cystectomy (16%) or TURBT followed by radical cystectomy (4%). At a mean follow-up of 14 months, the recurrence and metastasis rates were about 9% and 4%, respectively in patients undergoing partial cystectomies[11]. Complete excision is crucial, as it offers the best chance for a cure and minimizes the risk of recurrence. The extent of surgery depends on the tumor's location and size, with careful consideration given to preserving surrounding tissues and organs[12]. In cases where surgical resection is not feasible due to tumor location or patient comorbidities, alternative treatment options may be considered. Chemotherapy has been explored, particularly for unresectable or recurrent IMTs, with variable success. The use of targeted therapies, especially ALK inhibitors, has shown promise in patients with ALK-positive tumors, leading to significant tumor regression in some cases[8]. Chemotherapy is recommended when IMT is multifocal, invasive or shows local recurrence[13]. It is often a combination of agents including methotrexate, cisplatin, vinorelbine, adriamycin, carboplatin and paclitaxel given with a view of achieving complete remission[14].

Radiation therapy can also play a role, particularly for patients with incomplete resection or those with tumors that are not amenable to surgery. It may help control local disease and reduce the likelihood of recurrence[15].

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