


CASE REPORTS

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# Bladder inflammatory myofibroblastic tumor: report of four cases

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## Abstract

**Background:** Inflammatory myofibroblastic tumor (IMT) of the bladder (BIMT) is a rare entity with non-specific symptoms and particular histological features. Though bladder location is very uncommon, it arises from the bladder submucosal stroma as a polypoidal growth and is easily mistaken clinically, radiologically and histologically with a malignant neoplasm. The main treatment is surgical resection. To our knowledge, few reports of this pathology have been duly informed in Latin America to date.

**Case presentation:** We present four case male patients evidencing bladder tumor initially treated with transurethral bladder resection, diagnosing BIMT. They had shown tumor recurrence and laparoscopic treatment were decided without evidence of new recurrence.

**Conclusion:** Due to the few reports of this pathology, specially in South America, it is important to know its therapeutic management and follow-up.

**Keywords:** Inflammatory myofibroblastic tumor, TURB, Partial cystectomy

## 1 Background

IMTs are a rare soft tissue tumors. The first tumor was described in the lung in 1939 by Brunn [1]. Many different names have been assigned to this condition, including inflammatory pseudotumor, nodular fascitis, proliferation of pseudomalignant spindle cells, atypical fibromyxoid tumor, possibly attributed to the difficulty of differentiation due to its histological features [2].

The lung is the most frequent site of appearance, however, there are also reports of urogenital involvement. The first report of bladder involvement was described by Roth in 1980 [3].

The BIMT occurs more frequently in female patients (51.9%) with an average manifesting age of presentation of 38.9 years [4]. It generally comes attached with hematuria, increased voiding frequency, lower urinary tract

symptoms, dysuria, or hypogastric pain [5]. Surgical resection is normally their main treatment, and the role of adjuvant radiochemotherapy remains unclear.

We present the report of four cases of BIMT and the management carried out in each case.

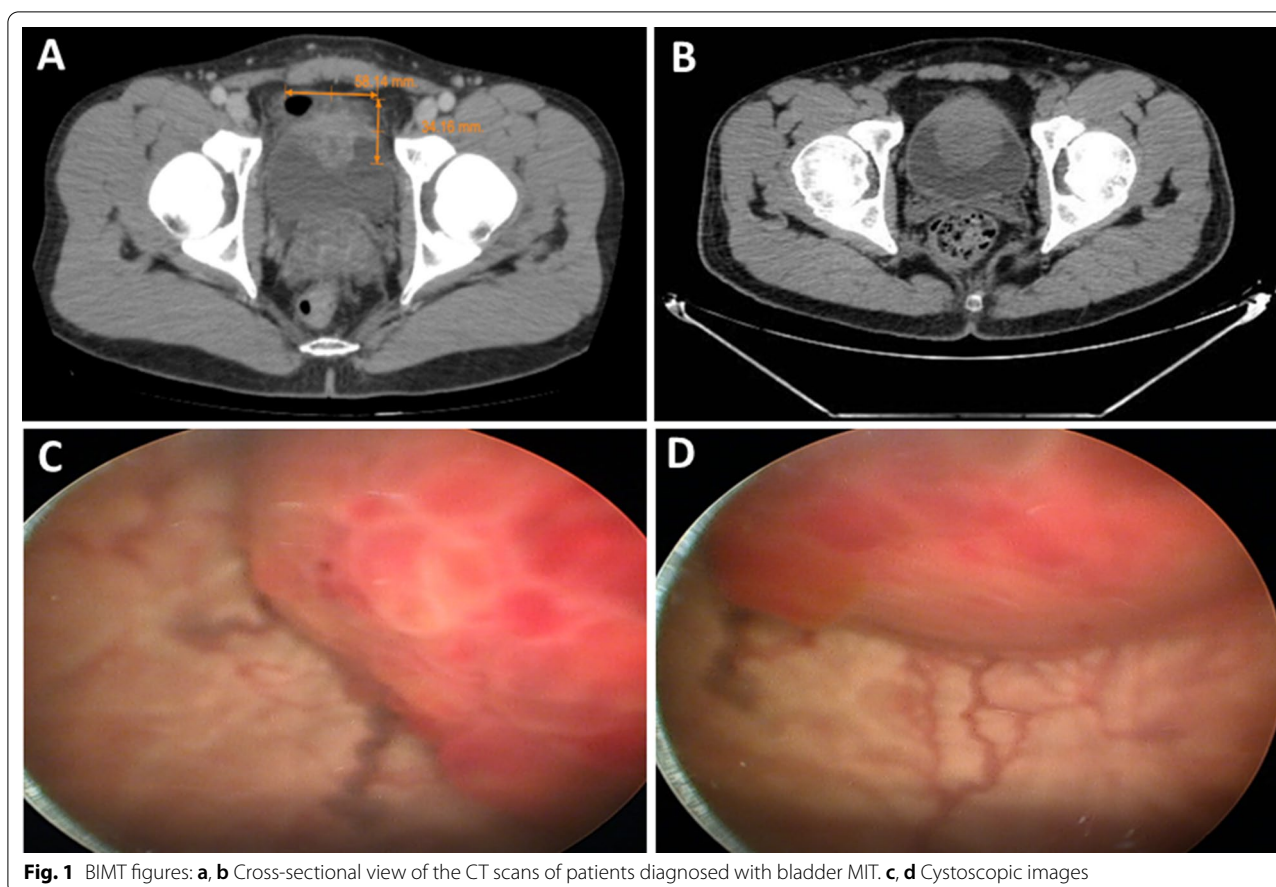
## 2 Case presentation

### 2.1 Case 1

A 36-year-old male patient, with no medical history, presented with lower urinary tract symptoms that started 2 weeks before consultation. He denies having had fever, dysuria, or hematuria. His urine culture was negative. Computed tomography (CT) scan showed at the level of the urachal implant, in the anterior sector of the bladder dome, a 41 × 34 × 58 mm expansive mass that enhanced heterogeneously after contrast administration, with extramural extension, which could be related to a primary urachus tumor (Fig. 1a). Preoperative cystoscopy examination showed a 5 × 4 cm solid tumor at the bladder dome.

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**Fig. 1** BIMT figures: **a, b** Cross-sectional view of the CT scans of patients diagnosed with bladder MIT. **c, d** Cystoscopic images

Incomplete transurethral resection of the bladder (TURB) was performed due to the tumor size. Microscopically, the submitted material showed spindle cell tumor with myofibroblastic cells and ovoid nuclei. They were arranged in poorly defined fascicles, with abundant cellularity consisting mainly with plasma cells, accumulations of xanthomized histiocytes, polymorphonuclear leukocytes, and eosinophils. Loose and edematous stroma with numerous congestive vessels. Atypical mitoses were not observed. The tumor was diagnosed as IMT.

Laparoscopic partial cystectomy was performed with the same pathological result, resection margins free of compromise. Immunohistochemistry report showed: Smooth muscle actin (alpha): Positive, ALK1: Positive.

Currently, the patient is disease-free after two years of surgery.

## 2.2 Case 2

An 18-year-old male, with a history of left cryptorchidism, complained of self-limited intermittent hematuria of 1 year associated with anemia. CT scan revealed a

46 × 43 mm sessile polypoid lesion on the right lateral aspect of the bladder with contrast enhancement and preserved perivesical fat.

A diagnostic cystoscopy informed a large polypoid formation measuring approximately 45 × 30 mm at the right lateral wall. TURB was performed. Pathology report informed inflammatory myofibroblastic tumor with microscopy of cells in poorly defined fascicles, intermixed with abundant cellularity consisting mainly in plasma cells, accumulations of histiocytes, polymorphonuclear leukocytes and eosinophils. Highly vascularized fibrous stroma, with bands of fibrocollagen tissue. Immunohistochemical revealed tumor cells stain positively for Smooth muscle actin (alpha), Desmin, and ALK1.

Three month after TURB, tumor relapse was evidenced by ultrasound. Cystoscopy confirmed tumor recurrence of 30 mm in the previous resection scar. Laparoscopic partial cystectomy was performed, the pathology report informed scar tissue with giantocellular granulomas and remnants of an IMT with free surgical resection margins.

Patient was followed for one year with no evidence of recurrence.

### 2.3 Case 3

A 38-year-old male, with no relevant history, presented hematuria for 15 days. No prior urologic disease history was found. Ultrasonography revealed a 60 mm × 40 mm heterogeneous mass at the bladder dome. Contrast-enhanced CT scan confirmed a solid-cystic mass in the bladder.

Complete resection by TURB was performed with a pathological report of an inflammatory myofibroblastic tumor. Microscopically revealed a pattern of abundant cellularity, spindle cells, histiocytes and vascularized stroma. Immunohistochemical staining revealed: Smooth muscle actin: focal positive, Desmin: focal positive, ALK1: Negative.

Six months later, he consulted for a new episode of self-limited macroscopic hematuria. Ultrasonography showed a 40 mm × 40 mm tumor recurrence in the bladder dome. Laparoscopic partial cystectomy was performed, with pathology confirming local recurrence, with free margins. No local recurrence has been detected after 3 years of follow-up.

### 2.4 Case 4

A 45-year-old man presented lower urinary tract symptoms associated with occasional episodes of self-limited haematuria for 2 months. No prior urologic disease history was found. Urine examination reported field covered with red blood cells, and ultrasound revealed a 40 × 48 mm hyperechoic mass at the bladder dome. CT scan showed a 61 × 59 × 55 mm voluminous solid

mass with wide implantation base, and transmural parietal involvement with apparent extravesical extension (Fig. 1b). A TURB was performed (Fig. 1c, d) with a histological report of spindle-shaped mesenchymal cells, areas of collagen, and mixed inflammatory elements with diffuse disposition in the lamina propria and the muscularis propria; immunohistochemically revealed the tumor cells positive for AML, ALK, AE1-AE3, CD 68 (Fig. 2).

Two months later, he presented a new episode of hematuria. The CT scan showed local disease recurrence, for which a laparoscopic partial cystectomy was performed with the same pathological result with free margins.

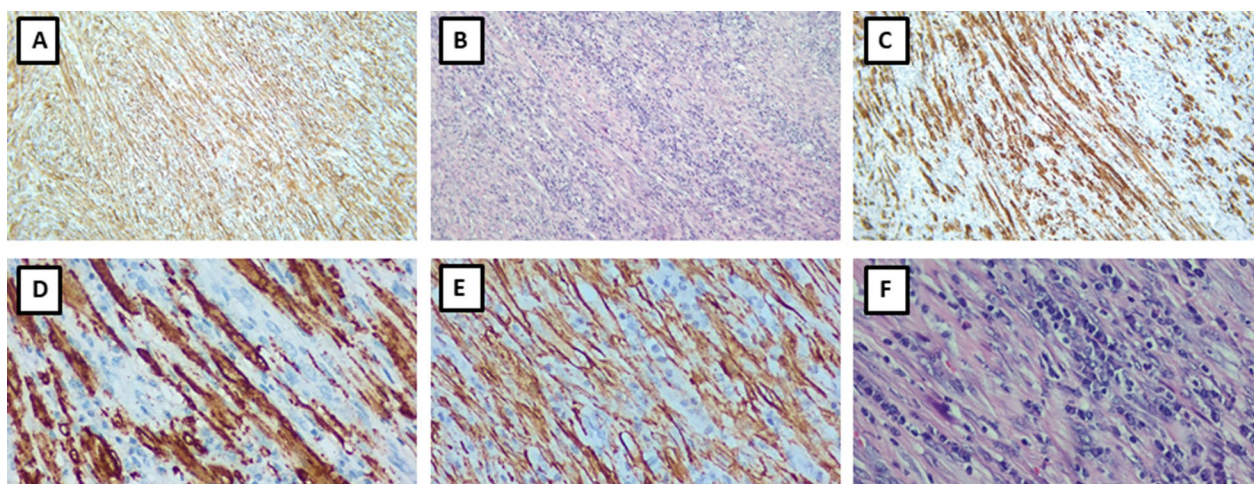
No recurrence or progression was observed during the 9-month follow-up time (Table 1).

## 3 Discussion and conclusion

The inflammatory myofibroblastic tumor is uncommon, composed by spindle and inflammatory cells, plasma cells and/or lymphocytes [6]. There is a lack of specific malignant features, such as anaplasia, pleomorphism, or atypical mitotic figures.

IMT shows immunohistochemical staining for vimentin, smooth muscle actin ( $\alpha$ SMA), calponin, cytokeratin AE1/AE3 and anaplastic lymphoma kinase (ALK). However, no significant differences were found regarding clinical presentation [4].

Until now, there are no known predisposing factors for its appearance in the urinary bladder. Etiology and pathogenesis still remain uncertain, but there may be an association with chronic infections such as Hepatitis B Virus, Epstein Barr Virus, Human Papilloma Virus [7]; bladder trauma, placement of polypropylene mesh (TVT) [8].



**Fig. 2** a, b H&E 10×–40×, Histological examination shows spindle cell proliferation with a fascicular pattern admixed with inflammatory cells on a myxoid background. Immunohistochemistry. c, d: AML diffuse positivity; e, f ALK diffuse positivity

**Table 1** Comparative chart

	Case 1	Case 2	Case 3	Case 4
Age	36	18	38	45
Sex	Male	Male	Male	Male
Clinical presentation	Low urinary tract symptoms	Hematuria	Hematuria	Low urinary tract symptoms + Hematuria
Tumor size by CT (mm)	41 × 34 × 58	46 × 43 × 40	58 × 40 × 42	61 × 59 × 55
Tumor location	Dome	Right lateral wall	Dome	Dome
1st Treatment	Incomplete TURB	TURB	TURB	TURB
Time to recurrence	–	3 months	6 months	2 months
Histological patterns	Spindle cells pattern	Mixed patterns	Spindle cells, Vascular pattern	Spindle cells, mixed inflammatory elements
Immunohistochemical staining	Smooth muscle actin, ALK1	Smooth muscle actin, ALK 1, Desmin	Smooth muscle actin, Desmin	AML, ALK, AE1-AE3, CD68

Rearrangements involving the ALK gene on chromosome 2p23 may also be involved. Therefore, the pathogenesis of IMT needs to be further studied.

Complementary examinations become relevant in BIMT for its diagnosis and follow-up. Typically, ultrasound, contrast-enhanced CT scan, and MRI can provide important information for estimating the size and involvement of the bladder wall.

Currently, surgical resection is the treatment of choice for BIMTs, such as TURB, partial cystectomy, and radical cystectomy, while some patients require reoperation due to recurrence, with a rate of 10–25% [9]. Comparatively, cystectomy is associated with lower reoperation rate [4]. Therefore, partial cystectomy is the first choice followed by diagnostic transurethral resection.

The therapeutic alternatives of endovesical instillation, ALK inhibitors, and embolizations are options that require further investigation and are not recommended yet.

IMT has a relatively good prognosis and is considered a tumor with intermediate biological behavior due to its low risk of metastasis, so far there are no reports of metastasis from BIMT [10].

Finally, the BIMT requires a periodic follow-up protocol after surgery, with images, urinalysis, and eventually cystoscopy.

#### Abbreviations

IMT: Inflammatory myofibroblastic tumor; BIMT: Bladder inflammatory myofibroblastic tumor; CT: Computed tomography; TURB: Transurethral resection of the bladder.

#### Acknowledgements

Not applicable.

#### Authors' contributions

V.B.R.F. contributed to conception, design, supervision, data collection, writing. M.M.M. contributed to data collection, analysis and interpretation, literature review. C.A.F. contributed to data collection, analysis and interpretation. C.J. contributed to analysis and interpretation, literature review. C.J.J. contributed to analysis and interpretation, literature review. J.J. contributed to supervision, writing. G.M.I. contributed to critical review. V.G.M. contributed to conception, design, critical review.

#### Availability of data and materials

The information was obtained from patients medical records.

#### Declarations

#### Ethics approval and consent to participate

All procedures performed in this study were in accordance with the ethical standards of our institutional review board with the 1964 Helsinki declaration and also approved by CEIAF (Comité de Ética de Instituto Alexander Fleming). Consent for surgical procedures and data storage were given by the patients.

#### Competing interests

Authors declare that there is no conflict of interest regarding the publication of this article.

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Received: 30 November 2021 Accepted: 3 February 2022

Published online: 08 March 2022

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