

## Case Report

# Inflammatory myofibroblastic tumor of the prostate

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### Abbreviations & Acronyms

ALK = anaplastic lymphoma kinase

IMT = inflammatory myofibroblastic tumor

MRI = magnetic resonance imaging

TURP = transurethral resection of the prostate

$\alpha$ SMA =  $\alpha$ -smooth muscle actin

**Introduction:** Inflammatory myofibroblastic tumors are borderline malignant soft tissue tumors primarily affecting the lungs and pelvic organs. This report presents a rare case of an inflammatory myofibroblastic tumor originating from the prostate gland in a young male.

**Case presentation:** A 20-year-old man developed gross hematuria and dysuria, revealing a prostatic mass. Pathological examination of a biopsy displayed spindle-shaped myofibroblast proliferation and an infiltrate of inflammatory cells, leading to a diagnosis of inflammatory myofibroblastic tumor. Following fertility preservation measures, the patient underwent a robot-assisted laparoscopic total prostatectomy with bilateral nerve sparing, resulting in a postoperative diagnosis of inflammatory myofibroblastic tumor. No recurrence was observed in subsequent imaging, and urinary continence was maintained.

**Conclusion:** Surgical resection appears effective in managing inflammatory myofibroblastic tumors of the prostate. This case underscores the importance of complete tumor resection due to the significant recurrence risk associated with inflammatory myofibroblastic tumors. Radical total prostatectomy emerges as a potential treatment strategy for prostate originating inflammatory myofibroblastic tumors.

**Key words:** inflammatory myofibroblastic tumor, prostate cancer.

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## Keynote message

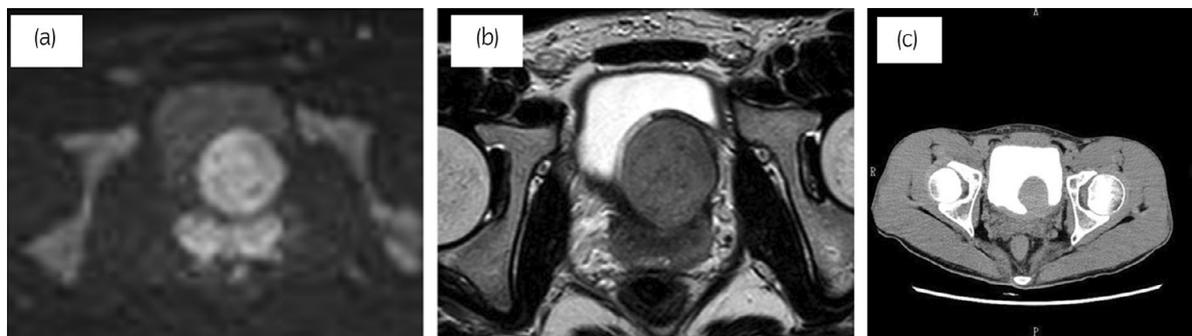
Inflammatory myofibroblastic tumors are borderline malignant soft tissue tumors that predominantly affect the lungs and pelvic organs and tend to occur in young patients. Although treatment has not yet been established, completed resection of tumor with radical total prostatectomy could be important due to the high recurrence rate of inflammatory myofibroblastic tumor.

## Introduction

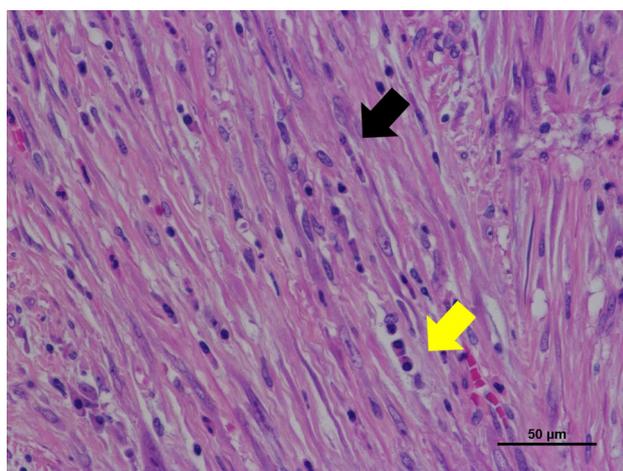
IMTs are borderline malignant soft tissue tumors that predominantly affect the lungs and pelvic organs and tend to occur in young patients.<sup>1</sup> In this report, we describe a case of an IMT of the prostate gland that developed at a young age.

## Case presentation

A 20-year-old man developed gross hematuria and dysuria, and a prostatic mass was found. He has no special family or medical history. There were no special findings in blood biochemistry tests. Prostate specific antigen was 0.717 ng/mL, which was within normal range. A contrast-enhanced computed tomography scan and a MRI showed a 35-mm-sized prostatic mass protruding into the bladder. MRI showed a prostate tumor protruding into the bladder, with a low intensity on T1-weighted images and a high intensity on T2-weighted images. The mass was close to the left ureteral opening (Fig. 1). He underwent transperineal needle biopsy of the prostate. Pathological examinations showed a proliferation of spindle-shaped myofibroblasts and infiltrating inflammatory cells, including lymphocytes and plasma cells (Fig. 2).



**Fig. 1** (a, b) Prostate mass with low signal on T1-weighted image and high signal on T2-weighted image. T1-weighted and T2-weighted images show a low-signal prostate mass. (c) Prostate mass in close proximity to the left ureteral opening is observed.



**Fig. 2** Proliferation of spindle-shaped myofibroblasts (black) and lymphocytes, plasma cells, and other inflammatory cell infiltration (yellow) such as lymphocytes and plasma cells.

Immunohistochemical staining showed weakly positivity for  $\alpha$ SMA, desmin, and ALK. Ki-67 staining showed that 30% of tumor cells were positive. He was pathologically diagnosed as an inflammatory myofibroblastic tumor (Fig. 3). After sperm cryopreservation for the fertility preservation, the patient underwent robot-assisted laparoscopic total prostatectomy with bilateral nerve sparing. Intraoperative findings showed a large prostatic mass protruding into the bladder. The patient underwent surgery while confirming the outflow of urine from the bilateral ureteral orifices, and the operation was completed without apparent complications. The postoperative pathological diagnosis was an IMT with negative margins (Fig. 4). Erection was confirmed on the fourth postoperative day. An MRI scan 7 months after surgery showed no evidence of disease on imaging and no urinary incontinence was confirmed.

## Discussion

Inflammatory myofibroblastic tumor is a disease marked by the excessive growth of spindle-shaped myofibroblasts and the infiltration of inflammatory cells, including lymphocytes

and plasma cells. The definitive diagnosis is made by pathological findings, which could be positive for  $\alpha$ SMA, ALK, and desmin by immunohistochemical staining, but the final diagnosis is based on morphologic findings. Previously recognized as a non-neoplastic lesion, it is now recognized as a borderline malignancy because distant metastasis and recurrence have been reported.<sup>2</sup> Surgical resection is the first-line treatment, and systemic therapy for advanced cases has not been established. This is the 15th case of inflammatory myofibroblastic tumor originated from prostate in the world and the second case in Japan.

Thirteen of the reported 15 cases underwent surgery: TURP in seven cases, radical total prostatectomy in four cases, and suprapubic prostatectomy in two cases. Four of the seven patients who underwent TURP experienced recurrence, while those who underwent radical total prostatectomy did not.<sup>3–5</sup> We chose radical total prostatectomy because of the high Ki-67 index (30%) and the high recurrence rate of cases with TURP. As a result, no obvious recurrence was observed 8 months after surgery, and maintenance the preservation of erectile function and urinary continence were achieved. Inflammatory myofibroblastic tumors in the prostate have been reported in very few cases, and there is no established treatment strategy but radical total prostatectomy could be considered.

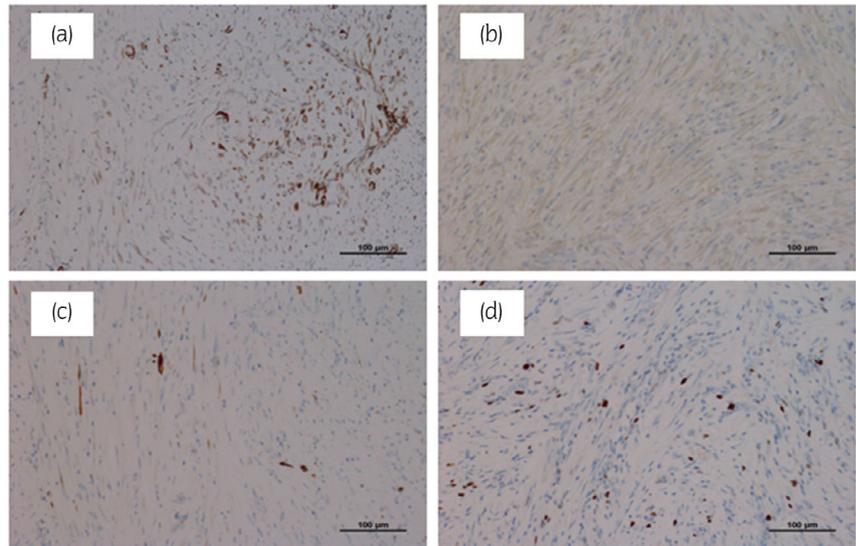
## Conclusion

We reported a young male patient with an IMT of the prostate.

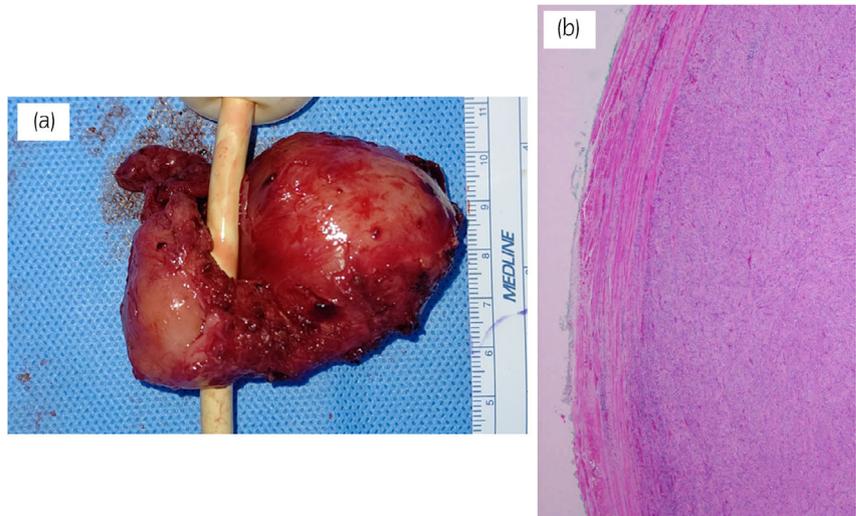
Completed resection of tumor with radical total prostatectomy could be important due to the high recurrence rate of IMT.

## Author contributions

Ken Fukiage: Writing—original draft preparation. Kazutoshi Fujita: Conceptualization and writing—review and editing. Shingo Toyoda, Mitsuhsa Nishimoto, and Takashi Kikuchi: Writing—review and editing. Takaaki Chikugo: Data curation. Kazuhiro Yoshimura, Atsunobu Esa, Akihiko Itou, and Hirotsugu Uemura: Supervision.



**Fig. 3** (a)  $\alpha$ SMA showing positivity in some cells. (b) ALK positive. (c) Desmin positive in some cells. (d) Ki-67 index is as high.



**Fig. 4** (a) Gross findings of excised prostatic tumor. (b) Microscopic image of the excised specimen tumor cells covered by the prostatic capsule are seen.

## Conflict of interest

The authors declare no conflict of interest.

## Approval of the research protocol by an Institutional Reviewer Board

Not applicable.

## Informed consent

Written informed consent was obtained.

## Registry and the Registration No. of the study/trial

Not applicable.

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