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CASE REPORT

Primary solitary fibrous tumour of the prostate: A case report and literature review

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Abstract

Introduction: Solitary fibrous tumour (SFT) is a rare mesenchymal tumour with intermediate malignant potential. Although this tumour arises in several sites, prostatic SFT is an extremely rare neoplasm and may prove confusing owing to the lack of clinical experience because of tumour rarity. The diagnosis may be further difficult because SFTs can manifest positive immunoreactivity for CD34 and progesterone receptor, which are known markers of prostatic stromal tumours. Herein, we describe a case of prostatic SFT that was difficult to differentiate from a prostatic stromal tumour of uncertain malignant potential because of positive immunoreactivity to CD34 and progesterone receptor. *Case Report:* A 40-year-old Japanese man presented with lower abdominal pain. Computed tomography revealed a prostatic mass; furthermore, prostate core needle biopsy revealed proliferating bland spindle cells, without necrosis or prominent mitoses. Tumour cells were positive for CD34 and progesterone receptor on immunohistochemical analysis; thus, a prostatic stromal tumour of uncertain malignant potential was initially suspected. However, as the tumour cells showed positive immunoreactivity for STAT6, the final diagnosis was an SFT of the prostate. The patient underwent tumour resection, and at the 6-month postoperative follow-up, neither local recurrence nor distant metastasis occurred. *Conclusion:* For an accurate diagnosis of an SFT of the prostate, STAT6 immunohistochemistry should be conducted for all mesenchymal tumours of the prostate. When STAT6 immunohistochemical analysis is unfeasible, pathologists should be aware that the morphological and immunohistochemical characteristics of SFT variable from case to case and diagnose with combined analysis of several immunohistochemical markers.

Keywords: prostate, solitary fibrous tumor, CD34, progesterone receptor, STAT6

INTRODUCTION

Solitary fibrous tumour (SFT) is a rare fibroblastic mesenchymal neoplasm.[1] The majority of SFTs are benign, despite being a mesenchymal tumour with intermediate malignant potential and a tumour that showed pulmonary metastasis has been previously reported.[2] SFTs can arise in different parts of the body, and extrapleural SFTs occur more frequently than pleural SFTs.[2] However, a prostatic SFT is an extremely rare lesion, and the epidemiology, pathology, and clinical characteristics of the disease are unclear. Here, we report about a case of prostatic SFT because of the necessity to differentiate it from a prostatic stromal tumour of uncertain malignant potential (STUMP) and also present a brief literature review.

CASE REPORT

A 40-year-old Japanese man with lower abdominal pain underwent computed tomography that revealed a prostatic mass, for which he was referred to our hospital, and a prostate core needle biopsy was performed. The biopsy specimen revealed bland spindle cells that proliferated in a sclerotic and fibrous background, without

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necrosis or prominent mitoses (FIG. 1a-c). On immunohistochemical analysis, the tumour cells tested positive for CD34 and progesterone receptor (FIG. 1d-e) but negative for desmin, smooth muscle actin, c-kit, DOG1, S100 protein, CDK4, and MDM2. The Ki-67 labeling index was approximately 1%.

Because of the positive immunoreactivity for both CD34 and progesterone receptor, we initially suspected a prostatic STUMP. However, a literature search revealed that SFTs could show positive reactivity for the progesterone receptor and that STAT6 is useful to distinguish STUMP from SFT of the prostate.[3] In this patient, tumour cells showed positive immunoreactivity for STAT6 (FIG. 1f), and thus, the final diagnosis was an SFT of the prostate. The patient underwent tumour resection 2 months after the diagnosis, and a solid tumour was resected from the prostate. The surgical specimen was fixed in 10% buffered formalin. Macroscopically, the tumour measured 60 × 50 × 40 mm. On cross-section, the tumour appeared as a well-defined, coarse, whitish gray, lobulated mass without necrosis or haemorrhage (FIG. 2a-b). Sections of paraffin-embedded tissue were prepared and stained with haematoxylin and eosin (HE) before examination using light microscopy. Histological examination revealed loose clusters of spindle cells (FIG. 2c). On immunohistochemical analysis, tumour cells tested positive for CD34, progesterone receptor, bcl-2, CD99, and STAT6 (FIG. 2d-f), and therefore, the tumour was diagnosed as an SFT of the prostate. At the 6-month postoperative follow-up, neither local recurrence nor distant metastasis had occurred.

DISCUSSION

Mesenchymal tumours of the prostate are extremely rare and are, therefore, often difficult to definitively diagnose. Prostatic stromal tumours, STUMP, and prostatic stromal sarcoma are the most frequently observed mesenchymal tumours.[4] However, a significant number of lesions can be included in the differential diagnosis including gastrointestinal stromal tumour (GIST), leiomyoma, leiomyosarcoma, rhabdomyosarcoma, schwannoma, malignant peripheral neuroectodermal tumour (MPNST), and SFT.[1]

Conventionally, SFT has been characterised by irregular distribution of tumour cells (pattern-less appearance) and a number of thin and branching vessels (staghorn appearance).[5] However, in practice, the morphological findings of SFT vary considerably from case to case.[6] Therefore, a morphological diagnosis based on the small specimen obtained from a needle biopsy of the prostate is rarely realistic, and immunohistochemistry proves essential for diagnosis. However, the results of immunohistochemistry should be cautiously interpreted because some immunohistochemical factors that are recognised as markers of SFT (e.g., bcl-2, CD34, CD99, progesterone receptor,

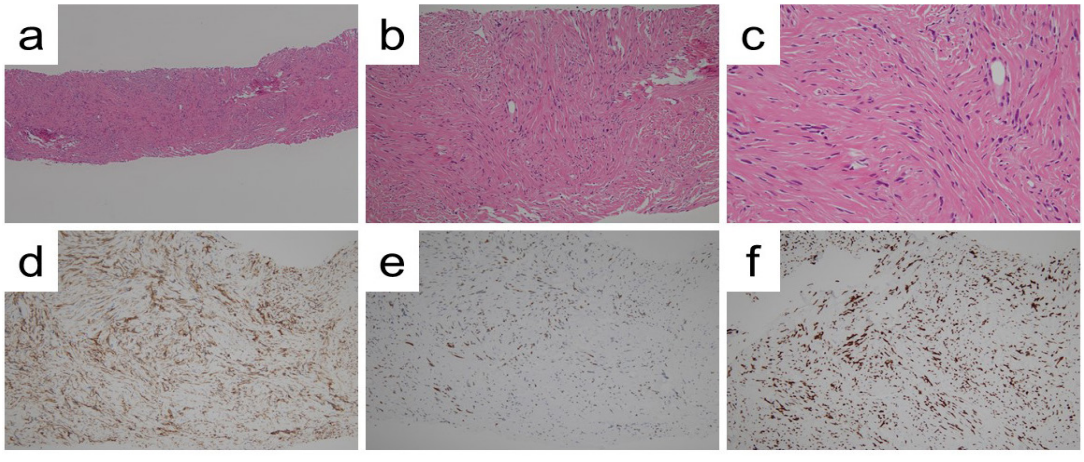

FIG. 1: Histopathological findings from the biopsy specimen. (a) Core needle biopsy was conducted for diagnosis (Hematoxylin–Eosin staining, ×40). (b and c) Proliferation of spindle cells arranged in a pattern-less state within a hypocellular collagen-rich background. In the biopsy specimen, a few branching vessels were found (staghorn appearance was indistinct; HE staining, ×100 and ×200, respectively). (d–f) Tumour cells showed positive immunoreactivity for CD34, progesterone receptor, and STAT6 (immunohistochemistry, ×100, respectively).

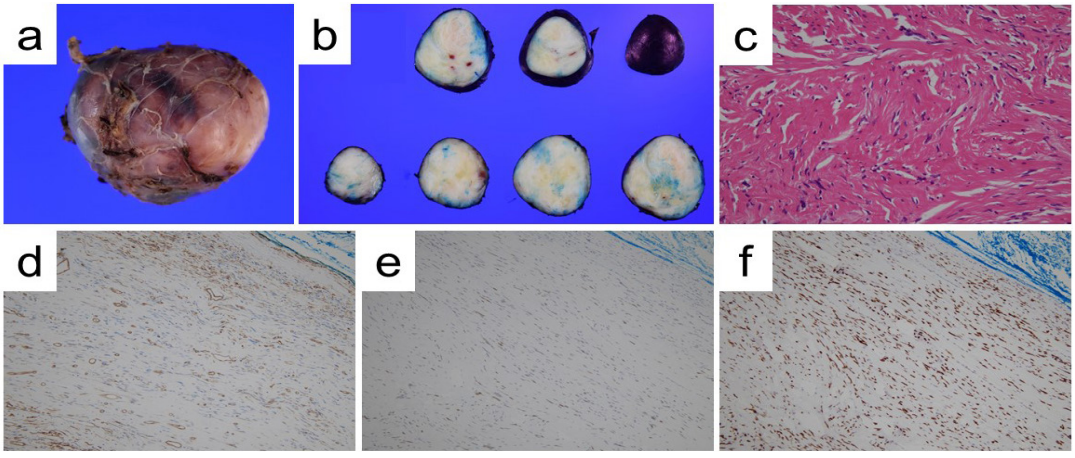

FIG. 2: Macroscopic and histopathological findings of the excised specimen. (a) The tumour measured 60 × 50 × 40 mm. On gross examination, a portion of the tumour shows a smooth-surfaced multinodular appearance. (b) On cross section, the tumour appears as a well-defined, whitish, lobulated mass without necrosis or haemorrhage. (c) Proliferation of spindle cells arranged in a pattern-less state within a hypocellular collagen-rich background in the biopsy specimen. Depending on tumour location, branching vessels were detected. (d–f) Tumour cells showed positive immunoreactivity for CD34, progesterone receptor, and

etc.) are not absolutely specific for SFT.

To elucidate the pathological characteristics of the SFT of the prostate, we undertook a literature survey of articles published up to April 12, 2020, in accordance with our previous algorithm.[7] We found 33 cases, and a total of 34 cases, including the present one, are summarised in Table 1. The mean age of the patients at the time of diagnosis was 59.1 (range, 36–87) years. The mean tumour size was 96.2 (range, 42–200) mm. Three cases showed direct invasion into the adjacent organs, such as the left piriformis

TABLE 1: Immunohistochemical staining of solitary fibrous tumour of the prostate

Immunohistochemical staining	Positive rate
ALK	0% (0/3)
Bcl-2	100% (23/23)
β-catenin	50% (6/12)
CD34	9.1% (34/35)
CD99	84.2% (16/19)
CD117 (c-kit)	9.1% (2/22)
Desmin	6.7% (1/15)
DOG1	0% (0/4)
Estrogen receptor	0% (0/6)
Ki-67	mean Ki-67 labeling index (n = 17) was 9.8 (range, 0–50)
Myogenin	0% (0/1)
Myoglobin	0% (0/2)
Pancytokeratins	4.0% (2/11)
Progesterone receptor	45.5% (5/11)
SMA	0% (0/18)
STAT6	100% (4/4)
S100	0% (0/13)
Vimentin	87.1% (88/101)

ALK, anaplastic lymphoma kinase; Bcl-2, B-cell lymphoma 2; CD, cluster of differentiation; DOG1, discovered on GIST1; STAT6, signal transducer and activator of transcription.

muscle[8], seminal vesicle[3], and urinary bladder.[9] One patient had multiple pulmonary metastases.[2] Based on the review of the identified cases, the immunohistochemical findings were determined as follows (the denominator varied for each immunohistochemical analysis because only papers describing positive or negative findings were analysed).

SFTs, exhibited the highest positive immunoreactivity rate for vimentin, bcl-2, and STAT6 (100%, 23/23, 10/10, and 4/4, respectively), followed by CD34 (97.1%, 34/35), CD99 (84.3%, 16/19), β-catenin (50%, 6/12), progesterone receptor (45.5%, 5/11), pan-cytokeratins (18.2%, 2/11), c-kit (9.1%, 2/22), and desmin (6.7%, 1/15). SFT had no immunoreactivity for smooth muscle actin (n=18), S100 (n=13), estrogen receptor (n=6), DOG1 (n=4), anaplastic lymphoma kinase (n=3), myoglobin (n=2), and myogenin (n=1). The Ki-67 labeling index (n = 17) was 9.8% (range, 0–50). These findings are summarised in Table 1.

These immunohistochemical findings from our literature review need further discussion. GIST is an important differential diagnosis and should be excluded before diagnosis of an SFT.[10] Morphologically, GIST differs from spindle, epithelioid, pleomorphic features, and SFT.[11] Furthermore, GIST often displays positive reactivity for CD34 and c-kit, but SFT could manifest positive immunoreactivity for both these markers.[3,11] Therefore, a histopathological diagnosis of a prostatic tumour with positive immunoreactivity for CD34 and c-kit should be undertaken with caution and should confirm STAT6 expression, as discussed further. If testing for STAT6 immunohistochemistry is not feasible, the confirmation of DOG1 expression is considered suboptimal to differentiate GIST from SFT because there are no reported cases of prostatic SFT with positive immunoreactivity for DOG1.

Furthermore, SFT must be differentiated from muscle tumours (leiomyoma, leiomyosarcoma, rhabdomyosarcoma, etc.).[1,12] As SFT has no immunoreactivity for muscle markers (SMA, desmin, myoglobin, and myogenin), except in a single case[13], a combination of immunohistochemistry for muscle markers might be helpful for a diagnosis. Schwannoma can be excluded through confirmation of S100 expression on immunohistochemistry. In contrast, some MPNSTs can show negative immunoreactivity for S100.[14] Thus, pathologists should confirm H3K27me3 loss in specific cases, as relevant.

It is most important to distinguish between SFT and prostatic stromal tumours, including STUMP. Despite high positive immunoreactivity to bcl-2, CD99, and vimentin, these markers have limited specificity as they are apparent in a variety of other mesenchymal tumours.[15] Therefore, it should be noted that tumour cells of SFT mainly showed positive reactivity for CD34 and progesterone receptor 97.1% and 45.5%. Both immunohistochemistry often show positive immunoreactivity for prostatic stromal tumours. Thus, an SFT of the prostate may be missed if the diagnosis is based on positive immunoreactivity for only CD34 and progesterone receptor.

We, therefore, sought further insight into the clinical usefulness of STAT6, which is a highly sensitive and specific nuclear marker for SFT.[3] Confirmation of STAT6 expression by immunohistochemistry can help differentiate SFT from other prostatic mesenchymal tumours.

In summary, to accurately diagnose an SFT of the prostate, STAT6 immunohistochemistry should be conducted for all mesenchymal tumours of the prostate. When STAT6 immunohistochemical analysis is not feasible, pathologists should be aware that the morphological and immunohistochemical characteristics of SFT variable from case to case and diagnose with combined analysis of several immunohistochemical markers.

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Authors' contribution: Yoichiro Okubo conceptualized the case report, integrated the data, and completed the manuscript as a major contributor; Suguru Nukada, Yosuke Shibata, and Kimito Osaka contributed to management of the patient and revised clinical description; Emi Yoshioka1, Masaki Suzuki, Kota Washimi, and Kae Kawachi carried out the histopathological evaluation and revised histopathological description; Takeshi Kishida contributed to management of the patient as a chief doctor of our hospital; Tomoyuki Yokose and Yohei Miyagi carried out the histopathological evaluation, integrated the data, and revised manuscript. Furthermore, all authors contributed towards the

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