Rapidly progressing malignant phyllodes tumor of the prostate with normal prostate-specific antigen levels: A case report

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ABSTRACT

Malignant phyllodes tumor of the prostate is a very rare entity. Here, we describe a 51-year-old patient with a malignant phyllodes tumor of the prostate with a poor prognosis and normal prostate-specific antigen levels. Digital rectal examination revealed a hard, nodular mass in the prostate, and magnetic resonance imaging exhibited a cystic mass measuring 8.7 cm × 7.0 cm × 6.7 cm. Immunohistochemical staining showed that the epithelial components were positive for CK8/18 and cytokeratin AE1/AE3; the atypical stromal cells were positive for CD34 and vimentin. Histological analysis resulted in a diagnosis of malignant phyllodes tumor of the prostate. Radical surgery was the treatment of choice. However, tumor recurrence was identified 6 months after the surgery, and the patient died 10 months after the surgery.

KEY WORDS: Immunohistochemical analysis, malignant phyllodes tumor, prostate, prostate-specific antigen

INTRODUCTION

Phyllodes tumors occur mostly in the breast but sometimes develop in the prostate.[1-3] According to its morphological classification, phyllodes tumor of the prostate is one of the four histological patterns of stromal tumors of the prostate with an uncertain malignant potential (STUMP).[4,5] Here, we describe a 51-year-old male patient who had a malignant phyllodes tumor of the prostate; in addition, we review previously reported cases.

CASE HISTORY

A 51-year-old man presented with a 2-month history of difficulty urinating. He showed no frequent urination, odynuria, or gross hematuria. Digital rectal examination revealed a hard nodule in the prostate that was not painful. Serum prostate-specific antigen (PSA) levels were within the normal range. Transabdominal ultrasonography showed a lobulated prostate tumour measuring 8.5 cm × 7.0 cm × 6.2 cm, which was indicated by multiple hypoechoic areas [Figure 1a]. Pelvic magnetic resonance imaging (MRI) revealed a hyperplastic nodule with solid and cystic changes with a dimension of 8.7 cm × 7.0 cm × 6.7 cm. Tumor invasion to the adjacent seminal vesicle was identified [Figure 1b]. An 8-core transrectal ultrasound-guided prostate biopsy was performed. Puncture pathology revealed that the lump surface was coated with elongated or slit-like glandular epithelium, but there was no evidence of adenocarcinoma; the spindle-shaped stromal cells exhibited high cellularity and a tangled pattern consisting of atypical cells with a smudged nuclear chromatin pattern without necrosis and showing a low mitotic rate (0–1/10 HPF) [Figure 2a]. Immunohistochemical staining showed that these atypical stromal cells were positive for CD34 and vimentin, but negative for estrogen receptor (ER), progesterone receptor (PR), androgen receptor (AR), p53, smooth muscle antigen (SMA), desmin, S-100, and CD117. The epithelial cells were positive for CK8/18 and AE1/AE3 and negative for PSA. The Ki-67 proliferation index was 15% [Figure 2b].

The patient agreed to resection of the prostate and the seminal vesicles at the Changhui Hospital (Shanghai, China). The mass surface was coated with benign epithelium, and significant hyperplasia
of atypical stromal cells was observed. Immunohistochemical staining showed that the stromal cells were positive for vimentin, CD34, and CD10, but negative for P504S, Bcl-2, PR, SMA, and S-100. The epithelial components were positive for CK8/18 and cytokeratin (CAM5.2) and negative for PSA. The Ki-67 proliferation index ranged between 20% and 30%. The pathological results were consistent with a diagnosis of malignant phyllodes tumor of the prostate with a negative surgical margin.

Tumor recurrence was identified 6 months after the surgery [Figure 3]. MRI revealed a hyperplastic nodule with solid and cystic changes measuring 9.1 cm × 9.5 cm × 10.2 cm, and two enlarged lymph nodes were found beside the left iliac vessel. The nodule occupied almost the entire pelvis and compressed the bladder, rectum, and left ureter. Serum PSA was within the normal range.

We then performed surgery with colostomy and nephrostomy of the left kidney. For induction therapy, the patient was treated twice with interventional embolization and radioactive particle implantation. However, after these therapies, MRI revealed progressive disease. The patient died 10 months after the surgery.

DISCUSSION

Phyllodes tumor of the prostate is a rare tumor that affects patients age 25–86 years (mean age, 55 years).[1! This tumor consists of a benign glandular epithelium and a mixture of variable cellular stromal components; it is classified as benign, borderline, or malignant, based on the pathological findings of stromal cellularity, cellular atypia, necrosis, stromal-to-epithelium ratio, and the number of mitotic figures on 1 × 10 high-powered field.[5! Previously reported phyllodes tumors were most commonly located in the female breast tissue, followed by the prostate.[6! Immunohistochemical studies showed immunoreactivity in glandular epithelium cells for PSA,[7! prostatic acid phosphatase, and AE1/AE3, and in stromal cells for CD10,[3! vimentin, and actin.[1! It has also been reported that these tumors often express ER and AR in epithelial cells,[7! thereby leading to the suggestion that antiandrogen agents may be useful for managing these patients.[8!"

Radical surgery is the treatment of choice for malignant phyllodes tumors of the prostate.[2! Complete surgical resection offers high rates of local control and disease-free survival.[6! The efficacy of radiotherapy and chemotherapy for malignant phyllodes tumors has not been well-established because of the rarity of these masses, although some reports have shown their efficacy.[3! Sakura et al.[9] reported the efficacy of VIP chemotherapy (VP-16, 100 mg/m²; ifosfamide, 1,200 mg/m²; and cisplatin, 20 mg/m² on days 1–5) for lymph node metastasis after prostatectomy in a 19-year-old man with a prostatic malignant phyllodes tumor. Murakami et al. reported the treatment of a 22-year-old patient with four cycles of VIP chemotherapy and eight cycles of AI chemotherapy (ifosfamide, 2,000 mg/m² on day 1 and doxorubicin, 30 mg/m² on days 1–3).[1! However, computed tomography revealed progressive disease, and the patient died 20 months after the surgery.[3!

In conclusion, we encountered a rare case of malignant phyllodes tumor of the prostate with a poor prognosis. Because the local recurrence rate of malignant phyllodes tumor of the prostate is extremely high, radical surgical resection provides the best chance for cure.[3!"
Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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