PAPILLARY CYSTADENOCARCINOMA OF THE PROSTATE : A CASE REPORT

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A 91-year-old man presented with nocturnal frequency and urge incontinence of a few days duration due to involvement of prostate cancer (PCa) accompanied by a large cyst in the left lobe of the prostate gland and urinary bladder wall. Channeling transurethral resection of prostate was performed to relieve the main symptoms and the resected material was histologically diagnosed as papillary cystadenocarcinoma arising from the epithelium of microscopic retention cysts. Following shrinkage of the large cyst, the patient is doing well on a combination regimen of a luteinizing hormone-releasing hormone analogue and bicaltamide. Papillary cystadenocarcinoma of the prostate was originally defined as papillary PCa arising from, not accompanied by, prostatic cysts. Cysts associated with PCa are subdivided into primary (or true) and secondary (or pseudo) cysts. Cancer cells in primary cysts originate from the epithelial lining. Papillary growth type cysts belong to this group and are regarded as papillary cystadenocarcinoma. The secondary (or pseudo) cysts, which have no epithelial lining and consist of hemorrhagic and/or necrotic contents are associated with invasive PCa. In the present case, the microscopic retention cysts revealed by histologic examination were of the primary type. This case of papillary cystadenocarcinoma, arising from a primary cyst, is the 13th such report from among previously reported cases in Japan.

(Hinyokika Kiyo **53** : 67–70, 2007)

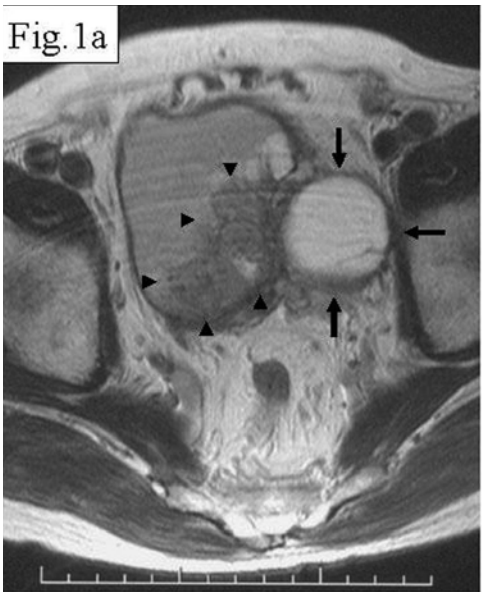
Key words : Papillary cystadenocarcinoma, Prostate

INTRODUCTION

Prostate cancer (PCa) associated with cysts is uncommon. Papillary cystadenocarcinoma is a rare type of PCa associated with prostatic cysts. Originally, it should have been defined as papillary PCa arising from, but not accompanied by, prostatic cysts. The present case is a very rare type of papillary cystadenocarcinoma, arising from microscopic retention cysts, with large prostatic cysts.

CASE REPORT

A 91-year-old man presented with nocturnal frequency and urge incontinence of a few days duration, preceded by difficulty in urination for two years. A marked enlarged and stony hard mass was palpated in the left lobe of the prostate gland by digital rectal examination. Laboratory data showed mild anemia (red blood cell count $306 \times 10^4/mm^3$, hemoglobin 9.6 g/dl, hematocrit 28.9%), renal dysfunction (blood urea nitrogen 36 mg/dl, creatinine 2.0 mg/dl) and highly elevated serum prostate-specific antigen (PSA) level (325 ng/ml). Drip infusion pyelography revealed that the left kidney was non-functioning and that the urinary bladder had been shifted to the right. Retrograde urethrocystography also revealed right displacement of the urinary bladder and prostatic urethra. Magnetic resonance imaging (MRI) demonstrated an approximately 5 cm hemorrhagic cyst (TI weighted high intensity) accompanied by an irregular tumor involving the left side of the prostate and urinary bladder wall (Fig. 1a, b). Cystoscopy revealed extrinsic compression of left lateral wall around the nonvisible left ureteral orifice. The echo-guided, transperineal systemic needle biopsy sample of the prostate demonstrated a typical well differentiated acinar adenocarcinoma (Gleason score 3+3). Then, channeling transurethral resection of prostate was performed to treat difficulty in urination, revealing a multilocular cyst focally covered by villous mucosa. Pathological examination of the resected material disclosed a typical well differentiated acinar adenocarcinoma and papillary cystadenocarcinoma arising from the epithelium of microscopic retention cysts, occurring multifocally in the specimens (Fig. 2a, b). Immunohistochemical examination revealed that papillary cystadenocarcinoma was negative for high molecular weight cytokeratin (34β E12) and positive for PSA. Periodic acid schiff and alcian blue were negative. Maximal androgen ablation therapy in combination with an luteinizing hormone-relesing hormone analogue and bicaltamide was carried out intermittently for 41 months as the patient expressed little desire to undergo castration. PSA dropped to within the normal range. The patient is doing well with shrinkage of the large cyst and without recurrence or metastasis of disease, although the non-functioning left kidney did not recover.



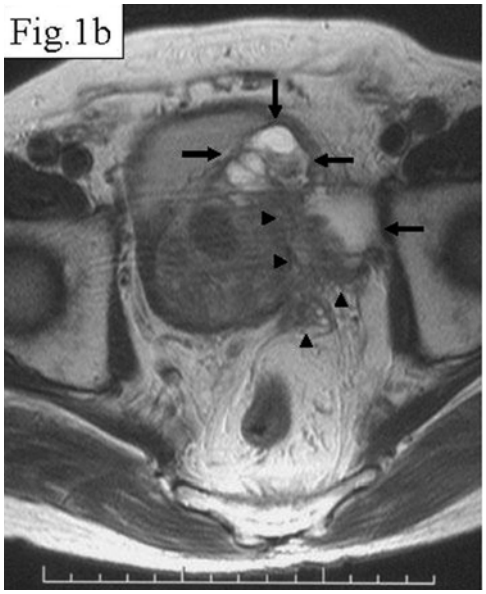
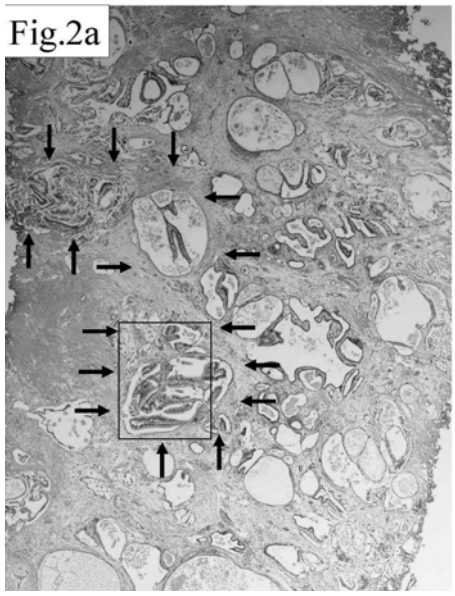


Fig. 1. MRI findings. (a) Large cysts (arrows) and irregular tumor (arrow heads) involving the left side of the urinary bladder. (b) Large cysts (arrows) and irregular tumor (arrow heads) involving the left side of the prostate gland.



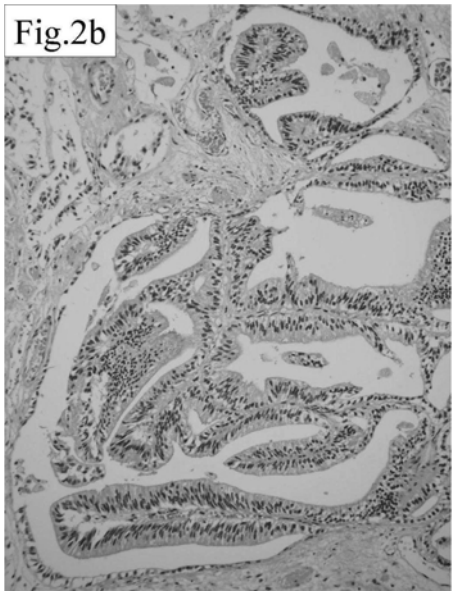


Fig. 2. Pathological findings. (a) Low power view of papillary cystadenocarcinoma (arrows) arising from microscopic retention cysts. Hematoxylin and eosin stain (original magnification, ×20). (b) High power view of black square (Fig. a). Papillary cystadenocarcinoma originating from epithelium of microscopic retention cysts and papillarily extending into the cystic space. Hematoxylin and eosin stain (original magnification, ×200).

DISCUSSION

Papillary cystadenocarcinoma of the prostate is a very rare type of PCa, although it is described in World Health Organization classifications. Originally, it should have been defined as papillary PCa arising from prostatic cysts, irrespective of congenital or acquired cysts[1]. Congenital cysts arise from remnants of Wolffian and Müllerian duct systems[2]. Acquired cysts are classified as simple retention cysts, and cysts associated with inflammation and PCa[2]. Although cysts associated with PCa are uncommon, they are subdivided into primary (or true) and secondary (or pseudo) cysts[1]. In the primary cysts cancer cells originate from the epithelial lining. Papillary growth type cysts belong to this group, and are regarded as papillary cystadenocarcinoma. The secondary (or pseudo) cysts, which have no epithelial lining and consist of hemorrhagic and/or necrotic contents are associated with invasive PCa. In the present case, the microscopic retention cysts revealed by histologic examination were of the primary type. Our case of papillary cystadenocarcinoma, arising from a primary cyst, is the 13th report among previously reported cases in Japan[3,4]. Patients with prostatic cysts usually present

between 35 and 55 years, with the clinical symptom of urinary obstruction[2]. The present case also had similar symptoms, possibly because the large cyst had compressed the prostatic urethra and urinary bladder. The treatment of prostatic cysts depends on the size and location. Thus some cysts may be amenable to endoscopic excision or drainage transurethrally. Aspiration of the cyst transperineally or transrectally under ultrasound guidance may also be a useful technique. Others may require open excision[5]. However, prostatic cysts associated with PCa usually shrink after the initiation of androgen ablation therapy except for those with hormone refractory PCa[6]. In the present case, the large cyst also shrank after the initiation of androgen ablation therapy, suggestive of a cyst arising from the prostate gland.

In 83% of the 12 cases previously reported in Japan[3,4], the disease was highly advanced at more than stage C so that they could not undergo radical surgery at clinical diagnosis. In such cases, hormonal therapy is common and is often effective for typical advanced stage PCa. Although 7 cases were treated by radical surgery, hormonal therapy was performed in only 5 cases of PCa with stage D2 disease. Two of the 5 patients treated by hormonal therapy, died of PCa at 3 and 69 months. Survival of the other three patients ranged between 24 and 53 months. However, the present case with stage C2 papillary cystadenocarcinoma was treated effectively with intermittent hormonal therapy with neither recurrence nor metastasis of PCa over 41 months despite the patient's age and advanced stage, suggesting that intermittent hormonal therapy could be a useful treatment modality for elderly patients with advanced stage papillary cystadenocarcinoma.

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(Received on February 6, 2006
Accepted on August 15, 2006)

和文抄録

前立腺乳頭状嚢胞腺癌の1例

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91歳，男性．主訴は数日前から続く夜間頻尿と尿失禁．PSA 325 ng/ml と高値で，前立腺の左葉が石様硬であった．DIP と UCG では膀胱と尿道は右側へ偏位し，左腎は無機能であった．MRIでは前立腺から膀胱の左側へまたがる径 5 cm の嚢胞を伴った腫瘍を認めた．前立腺生検では高分化型前立腺癌を認めた．排尿困難改善のためchanneling TUR を施行したところ，病理診断は貯留嚢胞上皮から発生した乳頭状嚢胞腺癌（Gleason score 6，C2）であった．内分泌治療は間欠的に施行し，治療開始後41カ月現在，嚢胞壁は縮小し，再燃・転移なく通院加療中である．嚢胞を伴う前立腺癌は比較的稀で，嚢胞壁が癌化する原発性嚢胞と癌が中心壊死・出血を起こし形成される二次性嚢胞に分類されている．自験例では原発性嚢胞と考えられる貯留嚢胞上皮が癌化した非常に稀な前立腺乳頭状嚢胞腺癌であり，本邦13例目であった．

（泌尿紀要 **53**：67-70，2007）