





Adenoid cystic/basal-cell carcinoma of the prostate following high-grade urothelial bladder cancer: a case report

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Abstract

Adenoid cystic/Basal-cell carcinoma (ACC/BCC) of the prostate is a rare histological type exhibiting various morphological characteristics and an optimal treatment has not yet been established. We report the case of a 63-year-old patient who complained of incomplete bladder emptying and recurrent urinary infection six months after transurethral resection of a high-grade urothelial bladder tumor. The clinical features, digital rectal examination, serum PSA levels, and multiparametric MRI did not refer to any suspicious prostatic lesions and cystoscopy revealed bladder neck hypertrophy, and yellowish zones in the prostatic urethra. Transurethral resection was performed due to these findings and histopathological analysis showed poorly differentiated ACC/BCC of the prostate. Even though there is no proven mutual correlation between ACC/BCC and urothelial bladder cancer, the appearance of obstructive urinary symptoms, bladder-neck hypertrophy, and macroscopic changes in prostatic urethra should be reconsidered for transurethral resection biopsy considering the possibility of ACC/BCC.

Keywords: adenoid cystic/basal cell carcinoma, prostate, urothelial carcinoma, bladder, case report

INTRODUCTION

Adenoid cystic/Basal-cell carcinoma of the prostate is a rare histological type accounting for approximately 0.01% of all prostate cancer cases first described in 1974 [1]. Since its various histologic and immunohistochemical features, outlining the diagnosis, treatment, prognosis, and outcome remains challenging, which results in delayed diagnosis and treatment and suggests poor prognosis. This type of prostate cancer is characterized by low serum PSA value since it arises from the basal cells and its diagnosis is incidental after transurethral resection of the prostate due to obstructive symptoms thus the stage when diagnosed is usually advanced. Even though the current treatment recommendation is primarily surgical resection, other treatment options may benefit the patients with continuous follow-up [2]. Most of the published case reports relate to the prostate but some cases report ACC of the urethra/Cowper's glands [3].

Even though there is no proven mutual correlation between ACC/BCC and urothelial bladder cancer, recently one case of ACC/BCC following urothelial bladder cancer was published [4].

We report an unusual case of a patient with high-grade urothelial bladder cancer followed by incidentally diagnosed early-stage prostatic ACC/BCC on the follow-up.

CASE REPORT

A 63-year-old male patient complained of recurrent urinary infections and a feeling of incomplete bladder emptying, otherwise in good general condition, without significant weight or appetite loss. Before six months he underwent transurethral resection of a non-invasive high-grade urothelial carcinoma (pTa) of the bladder without signs of tumor recurrence at a follow-up cystoscopy after three months.

The clinical work-up included a digital rectal prostate examination that did not refer to suspicious prostatic or periprostatic lesions. Prostate-specific antigen serum level was 0,657 ng/ml and MRI showed a preserved morphology and structure of the prostate with a preserved capsule and a diameter of 38.5×25mm with changes in the bladder wall due to previous transurethral resection of tumor but no pathological contrast accumulation

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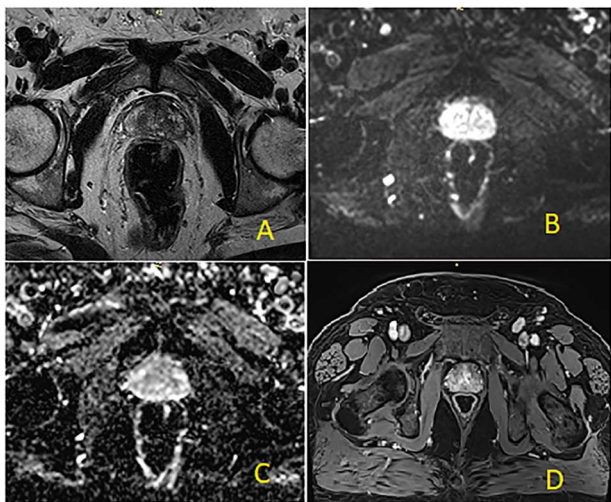


Figure 1. MRI findings in normal ranges A. T2-weighted imaging (T2WI), B. diffusion-weighted imaging (DWI), C. ADC map and Dynamic contrast enhancement (DCE), D. T1-weighted fast spoiled gradient echo.

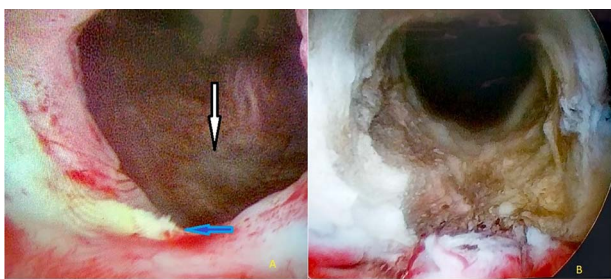


Figure 2. A. Bladder neck endoscopic visual, upper arrow pointing to the place of previously resected bladder tumor; lower arrow pointing to a new lesion of bladder neck before TUR, B. bladder neck and prostate after TUR.

was seen in the prostate tissue after administration of contrast or restriction of diffusion with PI-RADS score 1 (Fig. 1).

During the second follow-up cystoscopy which was performed six months after the resection of the urothelial bladder cancer, moderate bladder neck hypertrophy, and yellowish zones in the prostatic urethra were seen (Fig. 2A).

Due to the cystoscopy finding, obstructive symptoms, incomplete bladder emptying, and recurrent urinary infections, the patient underwent transurethral resection of the bladder neck, and prostate (Fig. 2B).

The histopathological report showed morphological features of poorly differentiated ACC/BCC of the prostate. The microscopic findings showed an infiltrative tumor composed of focally desmoplastic stroma infiltrated by nests of basaloid cells with hyperchromic nuclei and scant cytoplasm, showing peripheral palisading, admixed with adenoid-cystic and cribriform structures lined by a single to few layers of the neoplastic epithelium (Fig. 3).

Immunohistochemical staining for expression of CKHMW, p63, CK7, AMACR, GATA3, PAP, PSA, Estrogen, and androgen receptors was performed. The tumor was positive for CKHMW and CK7 (Fig. 4). The expression of p63 was partial, predominantly in the peripheral cells of the nests and adenoid-cystic structures and the PSA staining showed weak and partial signal with multifocal distribution (Fig. 5).

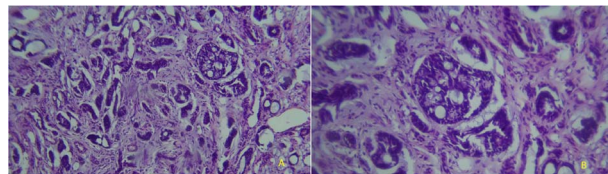


Figure 3. Basal-cell/adenoid cystic carcinoma; A. HeEo staining (40x) B. HeEo staining (100x).

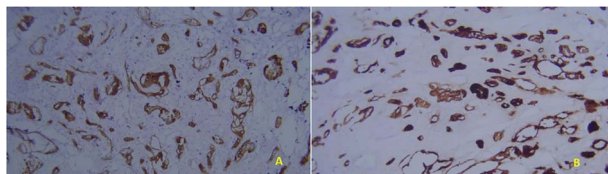


Figure 4. Immunohistochemical positive staining for; A. CKHMW (100x) B. CK7 (100x).

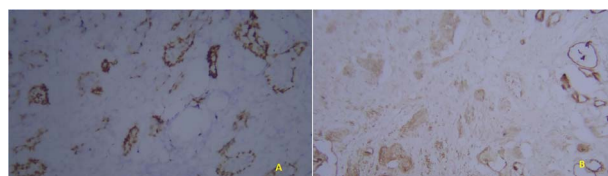


Figure 5. Immunohistochemical partially positive staining for A. p63 (100x), B. PSA (100x).

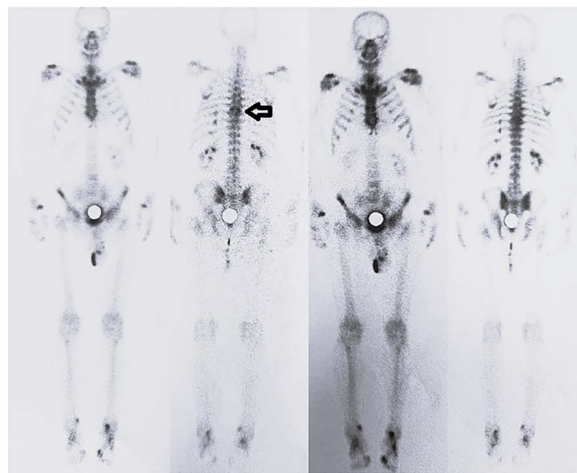


Figure 6. Skeletal scintigraphy with ^{99m}Tc -MDP showing the tracer's inhomogeneous distribution in the spinal column's thoracic segment, with emphasized activity in the projection of the eight thoracic vertebrae (arrow).

The immunostainings for AMACR, GATA3, PAP, Estrogen, and Androgen receptors were negative.

A metastatic workup was performed using a skeletal scintigraphy with $^{99\text{Tc}}$ -MDP showing the tracer's inhomogeneous distribution in the spinal column's thoracic segment, with emphasized activity in the projection of the eight thoracic vertebrae (Fig. 6). The patient had a history of vertebral injury, so the presence of metastasis in the thoracic vertebrae could not be reliably confirmed. According to the TNM classification for adenoid cystic prostatic carcinoma, the patient was in stage I (T1N0Mx).

The patient underwent definitive radiotherapy treatment in 38 fractions, with a total treatment dose of 70.4 Gy. The treatment

was carried out according to a controlled diuresis protocol, with the volumetric modulated arc therapy with Halcyon™ accelerator. Serum levels of PSA after radiation were 0,651 ng/ml. In further follow-ups, an enhanced abdominal CT scan and skeletal scintigraphy will be performed after 6 and 12 months.

DISCUSSION

The presented case is unique in presenting incidental diagnosis of ACC/BCC after transurethral resection of noninvasive high-grade carcinoma (pTa) in a patient with obstructive symptoms and normal serum PSA level. In the literature so far, there is only one reported case of a patient with high-grade muscle-invasive carcinoma, with incidentally diagnosed ACC/BCC [4]. In the presented case patient denied a family history of cancer and tobacco smoking was the risk factor for urothelial carcinoma.

Regarding the rarity of the ACC/BCC, the natural history of this tumor is not predictable. Case reports show various findings during clinical investigations such as digital rectal examination and ultrasonography, from an enlarged hard nodular prostate [5] to normal findings as in the case presented. Serum PSA and PAP levels are usually within the normal range. The most common diagnosis is incidental after transurethral prostate resection due to obstructive lower urinary symptoms [6].

The final diagnosis is based on immunohistochemical analyses, thus showing immunoreactivity for p63, CK7, and 34-beta-E12 stressing that CKHMW stains prostatic basal cells whose presence rules out usual-type prostatic adenocarcinoma [7], and distinguishes high-grade PIN (intact or fragmented basal cell layer) from adenocarcinoma [8].

A recent literature review showed that there is no uniformity in the management of such a rare prostate disease due to the lack of knowledge of its clinical and biological characteristics and the presentation of more cases is needed to overcome this challenge [1].

There are inconclusive data regarding the most effective systemic treatment and the effectiveness of androgen deprivation therapy, since ACC/BCC has demonstrated independence from androgen stimulation [9].

ACC/BCC in more than half of the cases has a poor prognosis due to prominent perineural and periglandular invasion suggesting aggressive behavior and has high-risk pathologic features or local recurrence, especially in younger patients [10].

Even though there is no proven mutual correlation between ACC/BCC and urothelial bladder cancer, the appearance of obstructive urinary symptoms, bladder-neck hypertrophy, and macroscopic changes in prostatic urethra should be reconsidered for transurethral resection considering the possibility of ACC/BCC.

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None to mention.

SUPPLEMENTARY MATERIAL

Supplementary material is available at the *Journal of Surgical Case Reports* online.

CONFLICT OF INTEREST STATEMENT

No conflict of interest.

FUNDING

None.

ETHICAL APPROVAL

The publishing of the case report was approved by the Ethical Board of the institution where the patient was referred.

CONSENT

A written consent of participation and publishing was obtained from the patient.

GUARANTOR

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