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CASE REPORT

Metastatic Adenoid Cystic Carcinoma of Prostate: Is Androgen Deprivation Therapy Beneficial?

[1]Shrawan K Singh, [2]Madhusudhanan Gnanasekharan, [3]Santosh Kumar, [4]Raguram Ganesamoni, [5]Amanjit Bal

ABSTRACT

Adenoid cystic carcinoma of prostate is a rare malignancy arising from basal cells of prostatic acini. The management of metastatic adenoid cystic carcinoma of prostate is not well defined because of its rarity. We report a 68-year-old male presented with acute urinary retention and pain in left hip region. Digital rectal examination revealed a hard nodular prostate with extension to the lateral pelvic walls. His serum PSA was 0.15 ng/ml. Transrectal ultrasound (TRUS) guided prostate biopsy revealed adenoid cystic carcinoma. Bone scan showed metastasis in left acetabulum. He underwent channel transurethral resection of prostate and bilateral orchiectomy. He received palliative radiotherapy for left acetabular metastasis to control his pain. At 3 years of follow-up, the patient is doing well with no new metastasis. Hormone therapy is a viable option in patients with metastatic adenoid cystic carcinoma of prostate.

Keywords: Carcinoma, Adenoid cystic, Prostatic neoplasms, Prostate-specific antigen.

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INTRODUCTION

Adenoid cystic carcinoma (ACC) is a common pathology usually involving the salivary glands. But, ACC of the prostate is a rare variant of prostatic carcinoma, comprising only 0.01% of malignant tumors of the prostate.[1] It originates from the basal cells of prostatic ducts and acini. It has been reported in patients with age ranging from 28 to 78 years (mean age of 50 years).[2] The management of patients with advanced and metastatic ACC of prostate is not well defined because of the rarity of this tumor. We report a patient with ACC of prostate with skeletal metastasis.

[1]Professor, [2]Senior Resident, [3,5]Assistant Professor, [4]Urologist

[1,3,4]Department of Urology, Postgraduate Institute of Medical Education and Research, Chandigarh, India

[2,5]Department of Pathology, Postgraduate Institute of Medical Education and Research, Chandigarh, India

Corresponding Author: Shrawan K Singh, Professor Department of Urology, Postgraduate Institute of Medical Education and Research, Chandigarh, India, e-mail: shrawanksingh2002@yahoo.com

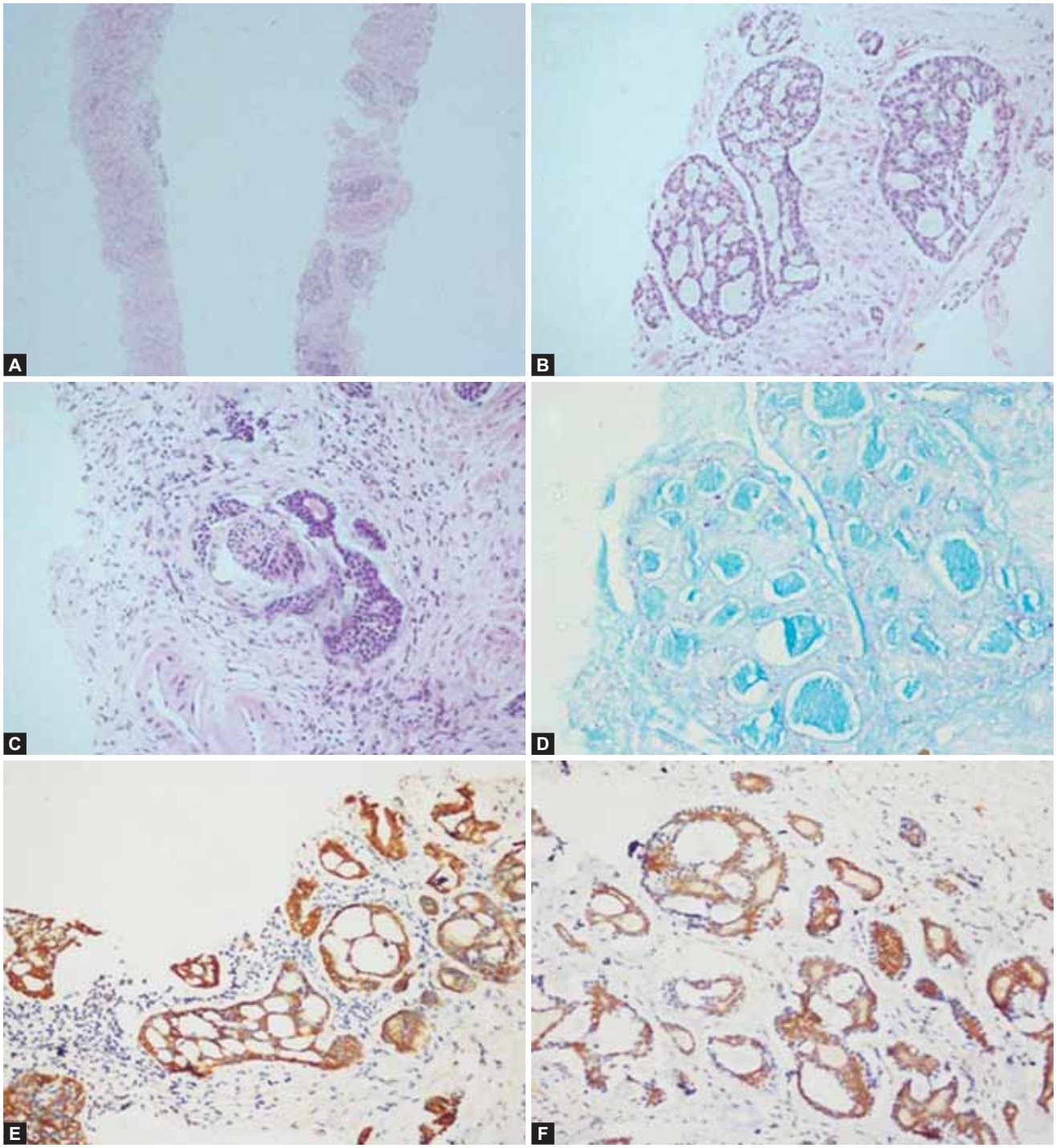
CASE REPORT

A 68-year-old male presented with obstructive lower urinary tract symptoms for 6 months and pain in left hip. He had undergone transurethral resection of prostate (TURP) in another hospital for similar complaints 3 years back. The histopathology report of the previous transurethral resection specimen was not available. Digital rectal examination revealed hard nodular prostate extending to lateral pelvic walls. His serum prostate specific antigen (PSA) was 0.15 ng/ml. Transrectal ultrasound-guided prostate biopsy showed tumor cells arranged in cribriform pattern with nests and columns of cells arranged concentrically around gland-like spaces filled with homogenous eosinophilic and granular basophilic material which was positive for Alcian blue stain. The cells were basaloid, small, round and angulated with hyperchromatic nucleus and scanty cytoplasm. Perineural invasion was noted. Foci of basal cell hyperplasia were also seen. Immunostains for PSA was negative while high molecular weight keratin was positive in both tumor as well as basal cell hyperplasia. Overall features were those of adenoid cystic carcinoma (Figs 1A to F). Immunostain for c-Kit and CK-7 were positive. Bone scan and X-ray pelvis revealed osteoblastic metastases in left acetabulum (Figs 2A and B).

He underwent channel TURP for relieving his bladder outlet obstruction. Though there is no established standard treatment for metastatic adenoid cystic carcinoma because of its rarity, there is evidence that basal cells of prostate might be responsive to testosterone though they do not produce PSA.[3] The patient underwent bilateral orchiectomy after discussion about its *pros* and *cons*. He also received radiotherapy to left acetabulum for hip pain which was not responsive to bisphosphonates. At 3 years follow-up, the patient is doing well with no new metastasis in bone scan.

DISCUSSION

Adenoid cystic carcinoma of the prostate arises from basal cells of prostatic ducts and acini and is composed of infiltrating basaloid cells forming dilated acinar and cribriform spaces with luminal basement-like material. Differentiation from basal cell hyperplasia and cribriform pattern of acinar adenocarcinoma may be difficult. The use of cytokeratin 34-β-E12 and prostate-specific antigen

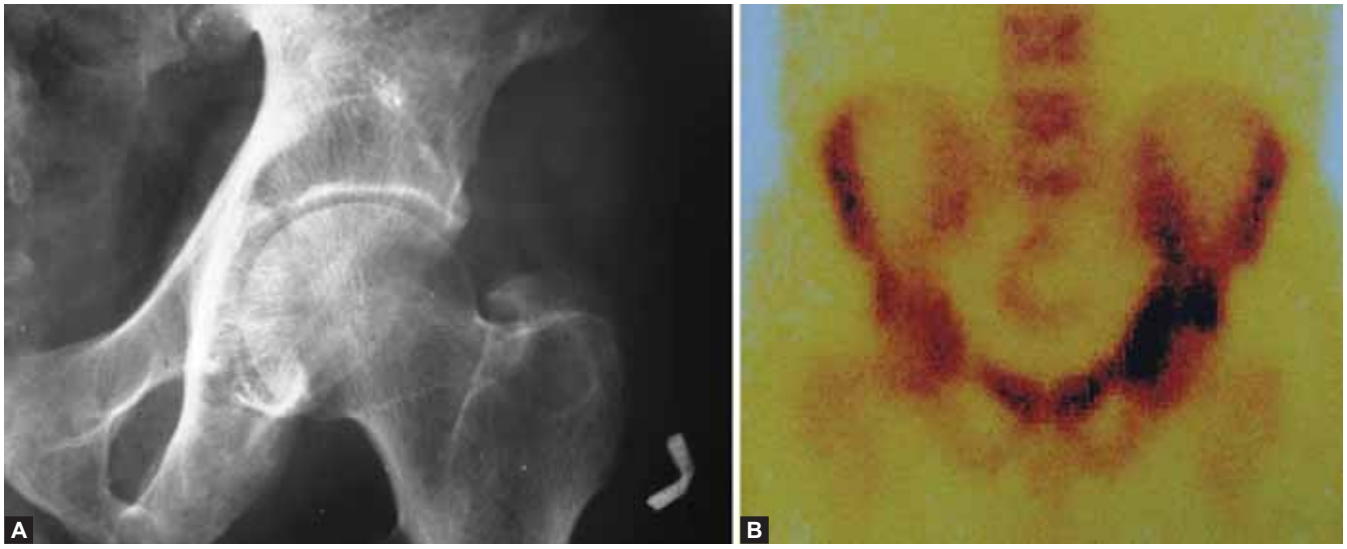


Figs 1A to F: (A) Adenoid cystic carcinoma involving the core in the right side (×20), (B) large cribriform units with lightly basophilic mucosubstance (×400), (C) perineural invasion (×400), (D) Alcian blue positive lightly basophilic mucosubstance (×400), (E) CK7 positive ductal epithelial cells (×400) and (F) c-Kit positive ductal epithelial cells (×400)

can help to differentiate from adenocarcinoma.[2] Elevated expression of Bcl-2 and higher Ki-67 proliferation index may aid in the differentiating from basal cell hyperplasia with prominent nucleoli.[4]

These tumors are malignant but slow-growing, and tend to spread locally via the nerves. The majority occur in the transition zone. Consequently, most patients present with symptoms of bladder outlet obstruction. The prognosis is better than prostatic adenocarcinoma. However, there is a small subset of patients in whom local recurrence and metastasis will occur. The metastases usually involve liver, lung and bowel but not bone, as is commonly observed in conventional prostate acinar adenocarcinomas.[5] Our patient developed skeletal metastasis which is rare. Radical surgery is the preferred first-line management option in localized carcinoma.[6]

These tumors do not respond well to chemotherapy or radiotherapy.[6] Hormonal therapy has not been studied



Figs 2A and B: (A) X-ray pelvis showing osteoblastic metastasis in left acetabulum and (B) bone scan revealing left acetabular

well previously. In a review of all previously published management strategies for ACC of prostate, Ayyathurai et al[6] found that only one patient had been managed with androgen deprivation therapy (ADT). However, the outcome was not reported for that patient. Since basal cells of prostate have been shown to have androgen receptors and testosterone responsive,[3] we did bilateral orchiectomy in our patient. The outcome in our patient was good since he had not developed any new metastasis at 3 years follow-up. Recently, Ahuja et al[7] reported the use of ADT in a patient with locally advanced ACC of prostate with stable disease in a limited follow-up of 6 months. Thus, out of the 50 odd patients with ACC of prostate in literature, our report is the first time in which a patient with metastasis had been managed with ADT with a longer follow-up.

Adenoid cystic careinoma (ACC) of prostate occupies one end of the spectrum of diseases arising from the basal cells of prostate, in which basal cell hyperplasia forms the benign end.[6] Due to limited number of reported cases with adequate follow-up, the prognosis and treatment options have not been clearly understood. In one series, metastasis has been documented in four of 15 patients who were followed up.[5] Therefore, a lifelong follow-up is necessary to detect local recurrence and metastasis.

CONCLUSION

Adenoid cystic carcinoma of prostate is a rare tumor whose clinical presentation is nonspecific, with the diagnosis generally obtained by transurethral resection or transrectal biopsy after the suspicion of an abnormal digital rectal examination. The treatment of choice for localized disease is radical prostatectomy because it is a potentially aggressive neoplasia; adjuvant treatment may be added. Hormone therapy is a viable treatment option in patients presenting with locally advanced and metastatic disease.

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