

**Case Report**

Primary Mucinous Carcinoma of Cowper' gland: A Case Report of a Rare Variant

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Abstract

Introduction: Primary carcinomas of the bulbourethral glands (Cowper's glands) are extremely rare.

Presentation of case: Herein, a 57-year-old man was presented by perineal mass for seven years. Physical examination revealed a nontender stony hard perineal mass without signs of inflammation. A urethrogram showed compression of the anterior part of the bulbous urethra. MRI of the mass revealed large perineal multilocular and marginal enhancement. The patient was managed by excision of the mass with safety margin. Histopathological examination of the mass showed remnants of malignant acini floating in pools of mucin which formed about 80% of tumor tissue. Immunohistochemical analysis revealed positive reactions of the tumour cells with cytokeratin 20 but negative reactions for PSA, β -catenin and cytokeratin 7.

Conclusion: We reported the first case of primary mucinous carcinoma arising in the Cowper's glands and the 22nd in the literature of Cowper's gland carcinoma.

Keywords: mucinous -carcinoma-Cowper' gland

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Competing Interests: The authors have declared that no competing interests exist.

Consent: We confirm that family members of the patients have given their informed consents for the case report to be published.

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Introduction

Primary carcinomas of the bulbourethral glands (Cowper's glands) are extremely rare, with only 21 reported cases in the medical literature and adenocarcinomas are the predominant histopathologic type in 17 of them [1]. Paquet and Hermann made note of the first example of this disease in 1884 [2]. The last case was reported in 2003 by Hitsamatu *et al* and the histopathological type was adenoid cystic carcinoma [3]. To the best of our knowledge, mucinous adenocarcinoma (MA) of Cowper's gland has not been reported in the literature. Herein we reported the first case of primary mucinous carcinoma arising in the Cowper's glands.

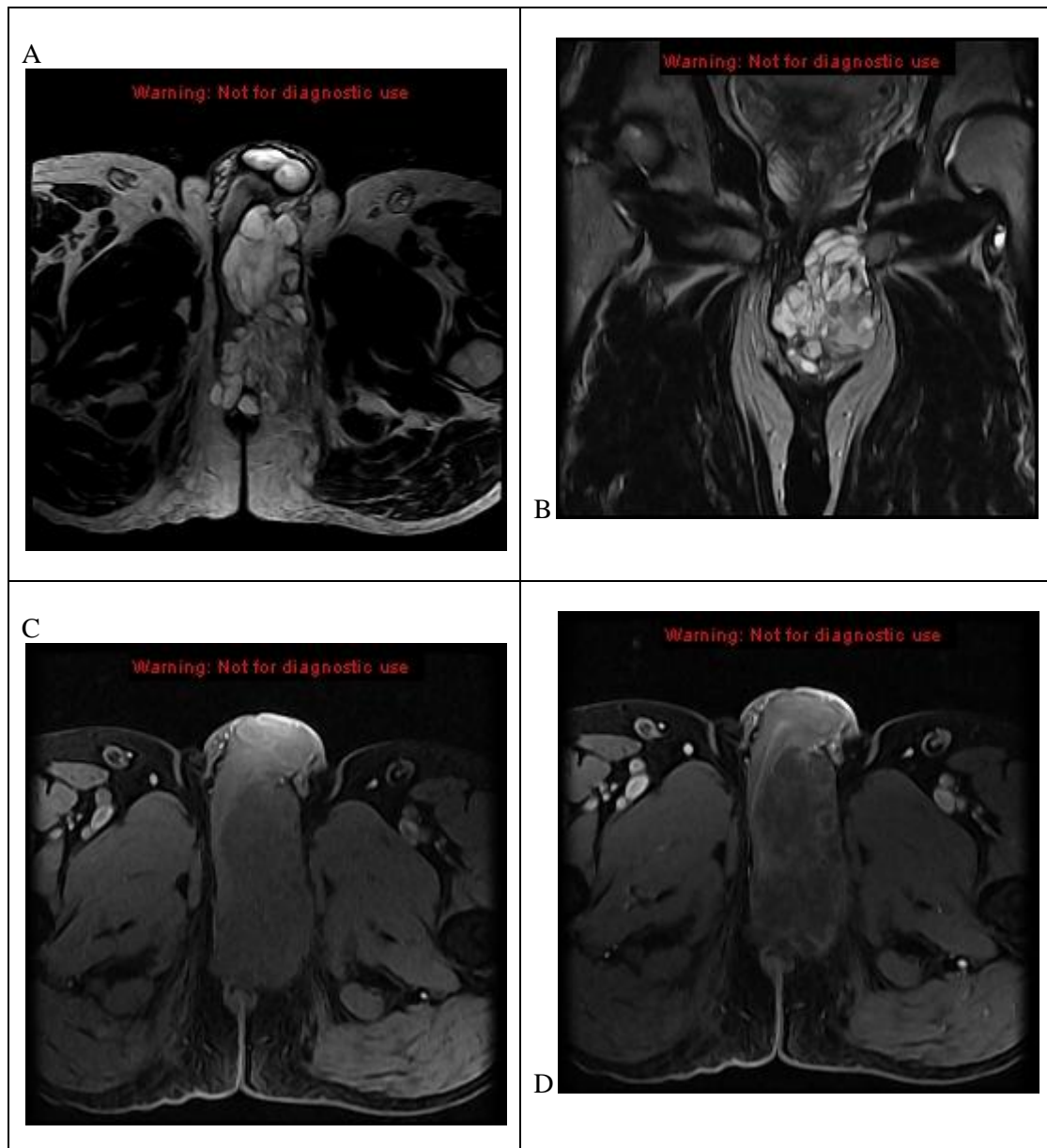


Figure 1 (A) axial spoiled gradient (SPGR) T1w MRI shows soft tissue mass at the left perineal region closely related to the penis displaying low SI with relative hyperintense septations, (B & C) Axial and coronal T2w MRI, the lesion displays high SI with hypointense septations (D) Gd-enhanced SPGR T1w shows mildly enhancing septations.

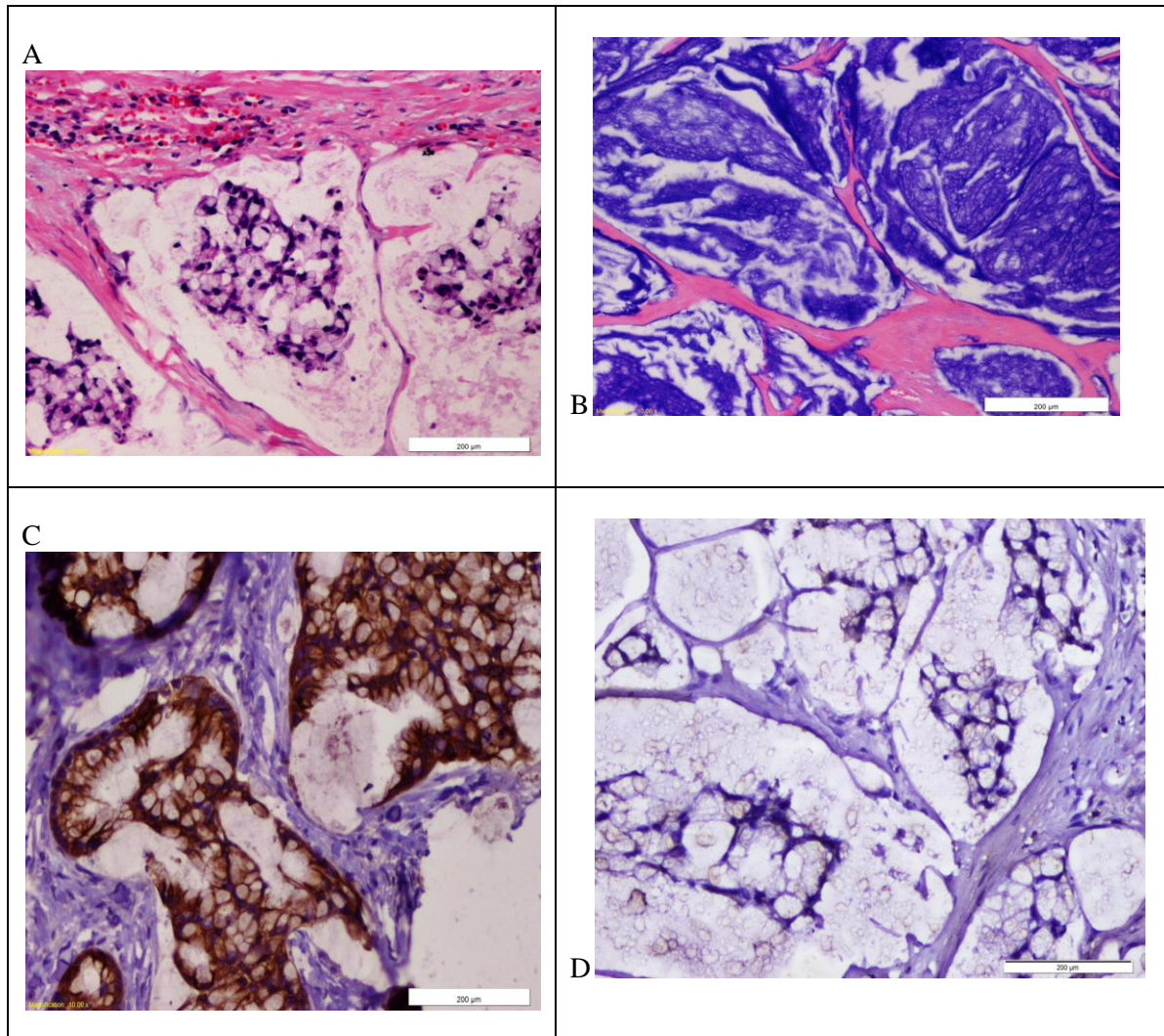


Figure 2 Couper's gland mucinous carcinoma A, showed remnants of malignant acini floating in pools of mucin (H&E stain x200). B, The mucin was positive for Alcian blue stain (x 200). C, the tumour cells showed positive cytoplasmic staining for CK20 (x200). D, The tumor cells showed negative nuclear staining for β -catenin(x 200)

Case presentation

A 57-year-old man was presented by perineal mass for seven years. He had no other urinary tract symptom. His past medical and family history were unremarkable. His serum creatinine was 1.1 mg/dl and PSA was 0.8 ng. Physical examination revealed a nontender stony hard perineal mass without signs of inflammation. A cystogram showed no abnormality. A urethrogram showed compression of the anterior part of the bulbous urethra. Cystourethroscopy showed a narrowing of the bulbous urethra which was interpreted as due to external pressure. The prostate, bladder and upper urinary tract were normal. MRI of the mass revealed large perineal multilocular and marginal enhancement. It is seen central and to the left invading the perineal root, the corpus spongiosum and left corpus cavernosum (figure 1). Primary rectal tumor was ruled out by colonoscopy. Microscopic analysis of the colon biopsy showed more or less normal mucosa and submucosa with

no evidence of masses or dysplastic change. The patient underwent Tru-Cut biopsy from mass which histologically showed only wide areas of mucin separated by fibrous tissues. After the biopsy, the patient was presented by a sinus discharging gelatinous material. The patient was managed by excision of the mass with safety margin. Macroscopic examination of the specimen revealed mass covered by elliptical piece of skin. The mass is about 10x6 cm, well circumscribed but not capsulated. The cut surface is greyish white with areas of cystic degeneration filled with gelatinous material. Histopathological examination of the mass showed remnants of malignant acini floating in pools of mucin which formed about 80% of tumor tissue (figure 2A) which was positive for alcian blue stain (figure 2B). Focal areas of signet ring components were also evident. All the safety margins were free. Immunohistochemical analysis revealed positive reactions of the tumour cells with revealed positive reactions of the tumour cells with cytokeratin 20 (figure 2C) but negative reactions for PSA, nuclear β -catenin (figure 2D) and cytokeratin 7. So, our final diagnosis was primary mucinous carcinoma of Cowper' gland.

Discussion

Cowper's glands are tubuloalveolar glands that are covered by a pseudo-stratified epithelium. Diseases affecting these glands are rarely identified; however, they can be host to infections, tumors, and congenital disorders [4]. Carcinoma of Cowper's gland is a very rare cancer [5]. The reported cases of Cowper's gland carcinoma have all been adenocarcinoma, and cystic adenocarcinoma [1]. However, mucinous carcinoma has not been reported. Herein we reported the first case of primary mucinous carcinoma arising in the Cowper's glands. This pathology is most commonly found in colorectal carcinoma (CRC). MA, a morphologic subtype of CRC, has more than 50% of the tumor composed of mucin, either extracellular with mucin lakes (colloid carcinoma) or intracellular where more than 50% of the tumor consists of signet ring cells (signet ring cell adenocarcinoma) [6]. In our case, extracellular mucin constituted about 80% of the tumor areas. So, the main differential diagnosis of the case is metastatic colon MA which closely mimics cowper gland carcinoma both morphologically and immunophenotypically. We excluded the possibility of metastatic colon carcinoma for the following reasons. First, the patient had not complained of any gastrointestinal problems. Second, the long history described by the patient (7 years) is not among the characteristic pathological features of colorectal MA as this tumor had worse clinical factors, including larger primary lesions, deeper invasion, higher rates of nodal and distant metastasis, and a larger number of metastatic sites [7]. Third, free colonoscopy. Recently, β -catenin has been reported to be of help in separating these tumors. Primary cowper's gland adenocarcinomas are usually negative for CK7 and nuclear β -catenin and positive for CK20 and CDX2 [8]. Our case was positive for CK20 and negative for CK7 and β -catenin which confirmed the diagnosis. The second important differential diagnosis is mucinous carcinoma of the prostate. The diagnosis of mucinous adenocarcinoma of the prostate rests on three criteria. First, at least 25% of the tumor should show aggregates of cells floating in lakes of extracellular mucin. Second, no signet ring component or significant intercellular mucin should be present. Finally, an extraprostatic primary must be ruled out. In addition, these carcinomas are positive for both PSA and PSAP, making immunohistochemistry a useful tool in ruling out extraprostatic primaries [9]. Our case had not been fulfilled these criteria as it showed signet ring components in addition to negative PSA staining.

Complete tumor excision is performed in the majority of patients [1]. However, pelvic exenteration was another line of treatment depending on each case and extension of the tumor. Recently, Cowper's gland carcinoma had been managed successfully by targeted therapy against C-Kit [10].

Conclusion

We reported the first case of primary mucinous carcinoma arising in the Cowper's glands and the 22nd in the literature of Cowper's gland carcinoma.

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