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Case Report

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MUCIN SECRETING ADENOCARCINOMA PENIS: A RARE TUMOR AT RARE SITE.

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ABSTRACT

Primary cutaneous mucinous carcinoma (PCMC) is a rare, slow growing, malignant neoplasm arising from the sweat glands. Though mucinous carcinoma is seen in various organs, primary mucinous carcinoma of the penis is a rare entity. More than 95% of the penile carcinomas are squamous cell carcinomas. We present a case of primary mucinous carcinoma penis, which, to best of our knowledge is the third case in the literature worldwide.

KEYWORDS: Penis, Mucin, Adenocarcinoma, Primary.

INTRODUCTION

Primary cutaneous mucinous carcinoma (PCMC) is a infrequent, slow growing, malignant neoplasm arising from the sweat glands. Lesions are most commonly seen on the head and neck and frequently mistaken for metastases from more common primary sites such as the breast, gastrointestinal tract, lung, ovary or prostate. First described by Lennox and colleagues in 1952, PCMC is also known as mucinous adenocystic carcinoma, primary mucinous carcinoma of the skin and colloid carcinoma.[1] PCMC involving penis is exceptionally rare, accounting for <5% of all penile carcinomas worldwide. This tumor is not included in WHO-2016 classification of tumors of male genital organs.[2] We report a case of 65 year old male who presented with non healing ulcer on penis which later was diagnosed as primary mucin secreting adenocarcinoma.

CASE REPORT

A 65 year old male presented with complaints of non healing discharging ulcer over penis for 9 months duration. Local examination revealed a 2 x 2 cm ulcer over dorsal aspect of shaft of penis(Fig.1A). There was no lymphadenopathy. All hematology and biochemical parameters were within normal limits. A provisional clinical diagnosis of squamous cell carcinoma was thought of. Biopsy taken from growth over penis shaft was sent for HPE.

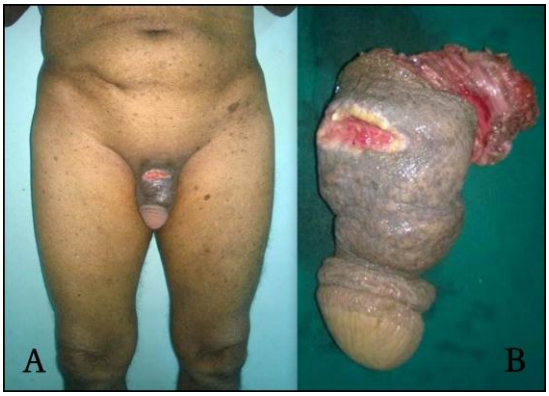

Figure 1

[**Figure1A**:Ulcer over dorsal aspect of shaft of penis.**1B**:Total penectomy specimen showing the ulcer].

The sections showed biopsy tissue lined by stratified squamous epithelium with elongation of Rete ridges and mild melanocytic proliferation. There was no evidence of dysplasia within the epithelium. In the deeper plane were seen large mucin lakes dissecting the muscle. In these mucin pools were seen floating glandular elements lined by columnar to cuboidal cells. Periodic acid Schiff(PAS) stain with diastase showed positivity in mucinous areas(Fig.2A,2B,2C). Diagnosis of mucin secreting adenocarcinoma ?primary ? metastatic was offered.

To rule out tumor in other common sites, CT and MRI of neck, thorax, abdomen as well as cystoscopy was done which revealed no evidence of tumor.

Total penectomy with perineal urethrostomy was performed which on HPE showed the presence of similar tumor.

The immunohistochemistry(IHC)showed positivity for carcino embryonic antigen (CEA) and cytokeratin (CK 7) and was negative for CK 20 and thyroid transcription factor (TTF 1),thus confirming the diagnosis of primary mucin secreting adenocarcinoma.(Fig.2).

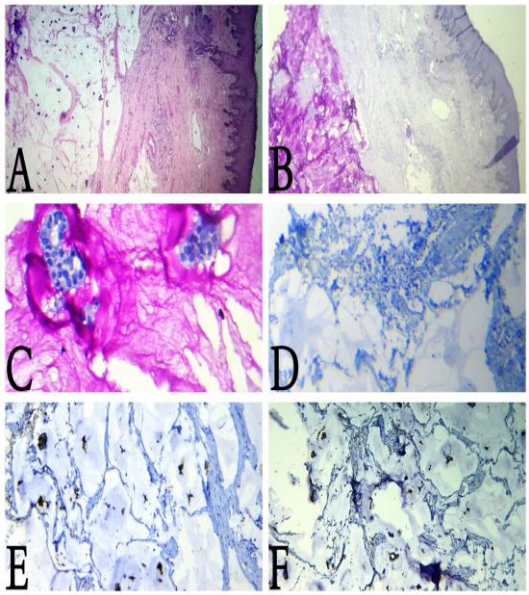

Figure 2

[**Figure 2A**:Photomicrograph showing normal squamous epithelial lining with tumor in deeper dermis.The tumor cell aggregates in glandular pattern embedded in lakes of mucin separated by thin fibrous septae. (H&E,x40**). 2B & 2C**: Special Stain-PAS Positive in mucinous areas.**2D**:IHC-TTF 1 Negative. **2E**:IHC-CK 7 Positive. **2F**: IHC-CEA Positive in tumor cells.

DISCUSSION

Nearly 95% of penile cancers are squamous cell carcinomas. Penile cancer usually originates in the epithelium of the inner preputium or glans. Proper histopathogenesis and etiological factors are to be elucidated but phimosis, balanoposthitis, balanitis xerotica obliterans, ultraviolet phototherapy,multiple sexual partners, early age of first sexual intercourse,a previous history of condyloma, poor penile hygiene, smoking and infection with Human Papilloma Virus (HPV) type 6,11,16 and 18 can cause penile cancer.[3]

The incidence of PCMC is about 0.07 per million and most often occurs during 6th to 7th decade of life.[3,4] Our patient was 65 years old, otherwise healthy male. To best of our knowledge this is the third case in the literature worldwide.[3,4] There have been case reports on mucinous metaplasia in penis ,which is considered to be a predisposing factor for penile mucinous cancer. The underlying mechanism of mucinous transformation remains unknown due to the limited number of cases.[5] We could not explicate the etiology in our patient.

On histology, PCMC is typically described as nests of epithelial cells floating in lakes of extracellular diastase-resistant, periodic-acid Schiff (PAS)-positive mucin.[6] Our case also showed similar findings(Fig.2B,2C). The IHC panel of CK (CK-7, CAM 5.2),CEA, epithelial membrane antigen(EMA), gross cystic disease fluid protein, S-100 protein, human milk factor globulins, alpha-lactalbumin show positivity while,TTF1 and 3 and estrogen and progesterone receptors status is variable.[7]

Our case showed positivity for CEA, CK 7 and negative staining for CK 20 and TTF 1.

Due to its rarity, cutaneous metastasis from more common sites of mucinous adenocarcinoma such as breast, lung, gastrointestinal tract, prostate and ovary, is often thought of. The final diagnosis of PCMC penis, therefore, can only be made after thorough clinical investigation to exclude a primary tumor elsewhere. IHC can also play a supportive role in the diagnosis. Recommended treatment includes wide local excision. Adjuvant hormone therapy with antiestrogenic agents, such as tamoxifen, help reduce the risk of recurrence. Recurrent PCMC appears to be resistant to both chemotherapy and radiation.[8] Our patient was followed up every month for first 6 months and then every 6 months to check for recurrence.

CONCLUSION

PCMC penis is extremely rare tumor. However, one has to think about it after exclusion of primary tumor elsewhere. Mucinous metaplasia can be the likely risk factor in its pathogenesis. Due to its rarity very scanty literature is available regarding its behavior and treatment protocol.

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