A case report on locally advanced squamous cell carcinoma in permanent perineal urethrostomy: A surgical conundrum!!

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ABSTRACT

Primary urethral cancer is a rare condition often manifesting in the sixth decade of life and is typically associated with urethral stricture disease. A 65-year-old male, a known case of stricture urethra, underwent perineal urethrostomy 10 years back. He presented with a growth at the urethrostomy site and the biopsy revealed squamous cell carcinoma. Magnetic resonance imaging of the pelvis revealed that the lesion extended into the penile and prostatic urethra with possible involvement of the corpus spongiosum, bulbospongiosus, and the bladder neck. Radical cystectomy, penectomy, scrotectomy, and bilateral orchiectomy with ileal conduit were performed. At 18-months of follow-up, the patient is recurrence-free.

INTRODUCTION

Primary urethral cancer (PUC) is a rare disease that usually manifests after the sixth decade of life and is more common in African Americans than Caucasian males.^[1] Preexisting stricture disease is present in at least 50% of the patients with a urethral carcinoma, however, squamous cell carcinoma originating in a perineal urethrostomy is seldom reported in the literature.^[2] Radical surgery with or without perioperative chemotherapy, chemoradiation, or radiation monotherapy are recommended treatment options for non-metastatic bulbar PUC. The benefit of more conservative surgical approaches must be weighed against the probability of local relapse or dissemination of the disease.

CASE REPORT

A 65-year-old Indian male, a known case of pan-anterior urethral stricture for 1 year, probably inflammatory in

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origin, had underwent a permanent perineal urethrostomy 10 years back.

The patient was asymptomatic until a year ago when an ulceroproliferative growth with foul-smelling discharge appeared at the site of the perineal urethrostomy. On examination, there was a fungating ulceroproliferative growth over the perineal urethrostomy site of 5 cm \times 5 cm involving the root of the penis and surrounding scrotal skin. Inguinal lymph nodes were not palpable, which was corroborated with a local ultrasound. Magnetic resonance imaging of the pelvis revealed a soft-tissue lesion in the antero-inferior aspect of the perineum with involvement of the penile and the prostatic urethra, corpus spongiosum, bulbospongiosus, and the bladder neck. A wedge biopsy confirmed the lesion to be squamous cell carcinoma and a 18-fluorodeoxyglucose positron emission tomography computed tomography (CT) scan was obtained which confirmed the absence of distant metastasis.

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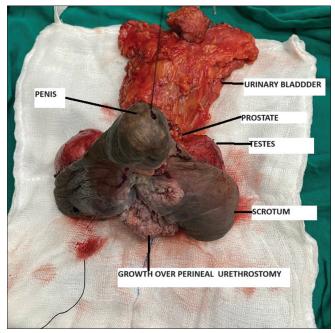


Figure 1: Postoperative en bloc specimen

The patient was planned for radical cystoprostactectomy with an ileal conduit and a standard pelvic node dissection along with total penectomy, scrotectomy, and bilateral orchiectomy via a combined abdominoperineal approach. A team of surgeons proceeded perineally by an inverted U-shaped perineal incision incorporating both the testes, the growth, and the penis with the apex in the mid perineum. A plane was created between the prostate and the rectum posteriorly, the levator muscles laterally, and the inferior pubic rami superiorly. Simultaneously through a lower midline incision, radical cystoprostatectomy with pelvic lymph node dissection was performed.

The specimen was delivered *en bloc* from the perineum and consisted of the bladder, seminal vesicles, prostate, penis, scrotum with the growth, bilateral testes, and pelvic nodes [Figure 1]. As the growth involved the root of the penis and scrotal wall, orchiectomy was performed to mitigate the risk of local recurrence.

Histopathology report showed a well-differentiated squamous cell carcinoma (pT4 N0 Mx) invading the dermis, corpus spongiosum, and cavernosum with adjacent scrotal skin involvement. No lymphovascular or perineural invasion was seen and all the margins were negative. Ten lymph nodes were present in the specimen and all were negative. At 18 months of follow-up, the patient is recurrence-free [Figure 2].

DISCUSSION

Dalbagni *et al.*, evaluated a cohort of 46 men with PUC treated primarily with surgery alone, and found that the 5-year overall survival for patients with posterior



Figure 2: Postoperative perineal photography after 1.5 years of follow-up

tumors was 26%, and only 4 of the 28 patients were free of disease. This observation led the authors to conclude that surgery alone is inadequate for advanced PUC.^[3] Later, Dayyani *et al.*, evaluated 44 men with advanced PUC and suggested that neoadjuvant cisplatin-based chemotherapy along with aggressive surgical resection is a viable option for high-risk cases and potentially offers a better prognosis in patients with bulbar tumors.^[4] Another treatment option is chemoradiation with external beam radiation to the external genitalia, perineum, and inguinal lymph nodes along with 5-fluorouracil + mitomycin and is known to be an effective treatment option for patients with advanced PUC.^{[5].}

All the patients reported in the literature had primary urethral carcinoma. The uniqueness of this case is the site of the urethral carcinoma which, was the perineal urethrostomy. In our case, due to foul-smelling discharge and a fungating growth, surgical intervention was favored over chemoradiation or neoadjuvant chemotherapy to mitigate the risk of septicemia and prevent surgical delay. The patient remains recurrence-free at 1.5 years of follow-up, without the need for adjuvant therapy and is currently on surveillance, however, long-term follow-up is requried due to the aggressive nature of the disease.

Urethral carcinoma is an uncommon and aggressive malignancy, underscoring the importance of early detection. Challenges in its treatment arise due to the rarity of the disease and the lack of standardized management.

This case highlights the central role of radical surgical resection in local disease control. Global reporting of such cases is essential for establishing consistent treatment guidelines through international collaboration.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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