LIBYAN INTERNATIONAL JOURNAL OF ONCOLOGY

Case report

Urothelial Carcinoma in Female Urethral Diverticulum: A Case Report

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Citation: Ben Omran A, Abusamra R, Abushnaf A, Alnari J. Urothelial Carcinoma in Female Urethral Diverticulum: A Case Report. Libyan Int J Oncol. 2024;3(2):64-66.

Received: 24-07-2024
Accepted: 23-10-2024
Published: 25-12-2024

Funding*: This research received no external funding.*

Conflicts *of Interest: The authors declare no conflict of interest.*

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Abstract

Female urethral diverticulum carcinoma (UDCs) is extremely rare. Only 126 cases of urethral diverticulum carcinomas in females have been documented. These tumors are usually diagnosed late, which worsens their prognosis. Because of its rarity, there is a significant variation in its management. We report another case of UDC in a 43-year-old Libyan woman who presented with recurrent UTIs.

Keywords. *Urethral Diverticulum Carcinoma, Female Urethral Diverticulum, Urothelial Carcinoma.*

Introduction

Urethral diverticulum carcinoma (UDC) is rare, of all female carcinomas it is account for less than 0.02% [1,2]. Transitional (urothelial) cell carcinoma accounts for 33% of these cases, squamous cell carcinoma and adenocarcinomas, account for 35% and 33% of female urethral diverticulum cancers, respectively [3]. Patients typically complain of hematuria and difficulty voiding; a palpable sub-urethral lump is the most typical symptom [4]. UDC has a variety of treatment options, local excisions as well as more aggressive treatments including radiation, chemotherapy, or anterior exenteration. Treatment failure occurred in 73% of patients receiving local excision only, resulting in either distant metastasis or local recurrence [5].

Case presentation

A 43-year-old white female which previously well, married and had three daughters, presented with a history of repeated hospital visits due to recurrent urinary tract infection (UTI) for three-month duration. She had no history of chronic disease. Physical examination revealed an anterior vaginal wall small mass. Urine routine examination showed microscopic hematuria. A Contrast-Enhanced Computed Tomography (CECT) showed urethral diverticulum 4cm in size with enhancement of small soft tissue mass within the diverticulum, there were multiple enlarged left peri-vesical lymph nodes. Diagnostic urethro-cystoscopy showed posterior urethral wall diverticulum containing papillary growth. Multiple biopsies were taken. The histopathology results revealed a high-grade urothelial carcinoma. We offered anterior pelvic exenteration followed by adjuvant therapy. Unfortunately, the patient refuses this option and asked for bladder preservation therapy. Transurethral resection was done, followed by EBRT and cisplatin-based chemotherapy. Her first follow-up CT scan showed a significant local recurrence with a new enlarged pelvic and para-aortic lymph nodes. A CT-guided lymph node biopsy was performed which revealed cancer metastasis. A three-doses of gemcitabine and cisplatin-based combination was given and after completion of this combination, she presented with a recurrent episode of gross hematuria and difficulty voiding. Hematuria was controlled by endoscopic fulguration. The patient is currently on palliative care.

Discussion

UDC represents only 0.001% of female genitourinary tract malignancies [6,7] in 1951 Hamilton report the first case of UDC [8]. Only 126 cases of UDC have been documented in the international literature [9]. According to literature there are different histological types of UDC, Transitional cell carcinoma (TCC) represent the second most common type of UDC in female which is the histological subtype in our case [10,11]. There are many theories for

development of UDC: The first is that they result from changes in the periurethral gland by recurrent infection and blockage [12]. The second theory suggests a metaplasia development resulting from chronic inflammation [13]. The last theory is that the malignant changes occurred in the mesonephric duct or Gartner remnants [14]. In our case, no history of chronic infection, and the disease was symptomatic by dysuria only in two months before diagnosis. Our case was relatively younger (43 yrs old) than the reported median age of the disease. The literature reports that the incidence of UDC varies by age group and racial background. 53 years old is the median age for presentation [15]. According to the most recent study conducted in 2018 by Eabhann O'Connor et al., white women (as our case) account for 38% of cases, followed by black women (32%), Asian women (22%), and Hispanic women (8%). (9) The presentation of UDC is vague; symptoms such as dysuria, dyspareunia, post-micturition dribbling, and urine incontinence may be present. More frequently, urethral bleeding or hematuria (55%), voiding dysfunction (16%), introital mass (13%), urinary tract infections (13%), and localized pain (6%), are the initial symptoms of UDC. Most individuals have a painless lump of the anterior vaginal wall on examination [16,6,9]. As we see no specific presentation of DUC in female, so that it is easy to miss its diagnosis. The main complaint in this case was dysuria, other symptoms were mild may be because of relatively smaller size mass. Among the diagnostic modalities for UDC are urine cytology, which has been shown to be positive in 91% of cases and may be a helpful first screening test [17,18], we ask for urine cytology in suspected cases of bladder cancer, especially in those with irritative symptoms. A number of imaging modalities, including CT, MRI, voiding cystourethrography, intravenous urography, ultrasound, and cystourethroscopy, have been utilized to help diagnose UDC [19]. To check for bone metastases, distant tissue, and enlarged lymph nodes, a CT scan or an isotope bone scan may be used [20]. Because the periurethral glands are located inside the periurethral region that borders the paravaginal fascia, staging UDC is extremely challenging. In certain instances, the TNM staging system has been applied [21]. Most cases are at an advanced stage at diagnosis because there is a little muscle underlying them. They also tend to be high grade with 73% grade 2 or 3 at presentation [22,19].

Surgery is the cornerstone of care, and available options include urethral diverticulectomy (+/-pelvic or inguinal lymph nodes. +/- chemo-radiation). Anterior pelvic exenteration (removal of the bladder, uterus, anterior vaginal wall, and pelvic lymph nodes +/− adjuvant treatment). Radiotherapy +/− adjuvant chemotherapy alone [9]. More encouraging outcomes were shown with anterior exenteration combined with urine diversion (which was not accepted by our patient) as 87% of patients were disease-free after six months to two years [23].

Conclusion

UDC in female is extremely rare, and due to its non-specific presentation, it is usually diagnosed lately and patient presented in advanced stage. Since UDC is an aggressive disease, aggressive care is required. Up to now, the optimal treatment appears to be radical surgery, and a multimodality treatment approach seems to be the best approach to improved overall survival.

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