CASE REPORT

Case Report: A rare case of mucinous adenocarcinoma of the female urethra [version 1; peer review: 1 approved with reservations]

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First published: 13 Jun 2023, 12:664
https://doi.org/10.12688/f1000research.132303.1
Latest published: 13 Jun 2023, 12:664
https://doi.org/10.12688/f1000research.132303.1

Abstract

Background: Urethral adenocarcinoma is a very uncommon problem in women, with a prevalence of 0.02%. Due to the disease's rarity, there is very little information available about it. Treatment for cancer can have a significant negative influence on a patient's condition. Mucinous adenocarcinomas are a very uncommon type of cancer that is poorly studied. These cancers almost invariably have a worse outcome compared to conventional adenocarcinomas.

Case Presentation: A 67 year old woman presented with hematuria and stranguria. A computerised tomography (CT) scan of this patient revealed an urethral tumor. In her histopathologic report we found the tumor cells were arranged in small clusters and single cells were scattered among extracellular mucin and signet ring cell picture, which is consistent with mucinous adenocarcinoma. We then performed partial urethrectomy with negative surgical resection, but the tumor reappeared 18 months later with no symptoms. The patient then decided to undergo radiotherapy for 33 sessions. From a recent MRI follow up the patient remains recurrence free.

Conclusions: Early surgical treatment with or without adjuvant radiotherapy appears to be the best option in cases of small, organ-confined disease of urethral mucinous adenocarcinoma. Partial urethrectomy can be performed in this type of cases, which can prevent the use of permanent urinary catheters and further improve the patient's quality of life.
Keywords
Mucinous adenocarcinoma, partial urethrectomy, urethral neoplasm, radiotherapy

This article is included in the Oncology gateway.
Introduction

Urethral adenocarcinoma is a rare neoplasm that makes up for 0.02% of malignant tumors in females.1 There are only a few reports about this neoplasm in Indonesia. Mucinous adenocarcinomas are an extremely rare and poorly understood kind of cancer. These tumors have traits in common with a different class of tumors known as signet ring cell adenocarcinomas, which express mucin inside their cells. Compared to other adenocarcinomas, urethral adenocarcinoma frequently has a poor prognosis.2 The objective of this study was to share our experiences managing a female patient with mucinous adenocarcinoma of the urethra who had been monitored for four years.

Case presentation

A 67-year-old, retired single female patient from Indonesia presented with blood and clot gushing out from her urethra. She came with stranguria and hematuria. There was no previous history of urinary retention. The patient had a hysterectomy due to fibroids in 1991 and cholecystectomy in 2017. There was no previous history of cervical cancer in her family. She often consumed red meat and drank coffee. Her menarche history began at 12 years old and menopause was at 51 years old.

We performed a computerised tomography (CT) scan of the abdomen and suspected a urethral tumor, which was followed by a biopsy. We saw a tissue partially lined with locally hyperplastic transitional epithelial cells showing hyperplastic growth, forming a tumor mass with infiltrative growth from our histological examination. Tumor cells are arranged in small clusters and single cells are scattered among extracellular mucin, some of which form glandular structures. Tumor cells with round-oval nuclei, pleomorphic, hyperchromatic, vesicular, some with nucleolus, eosinophilic cytoplasm, some vacuolated. Tumor cells appear with a "signet ring cell" picture (Figure 1). The stroma is filled with mild to moderate acute and chronic inflammatory cells, with areas of bleeding. No tumor emboli were found in the vessels. It is consistent with mucinous adenocarcinoma.

Subsequently we performed an urethrocystoscopy examination. From our examination we can distinguish between healthy tissue and tumor border. We decided to perform a partial urethrectomy. From the resected tumor, it was found that the surgical resection margins were negative from neoplastic involvement, no lymphovascular invasion was identified. After the procedure the patient had been in remission. However, after 18 months, from Magnetic Resonance Imaging (MRI) examination, recurrence had appeared. From our physical examination, it showed a mass in the urethral area. The patient did not feel any symptoms. Then the patient underwent radiotherapy for 33 sessions. Currently, she remains free of recurrence for 22 months of follow-up (Figure 2). The MRI examination showed no residual mass was seen in the urethral area. No paraaortic, parailiac, and obturator lymphadenopathy was seen. No abnormalities were seen in other intra-abdominal and pelvic organs (Figure 3).

Discussion

Female urethral carcinoma is a condition with a modest prevalence, accounting for 5% of female urological tumor cases and 0.02% of all instances of malignant tumors in women.1,3 Mucinous adenocarcinomas are an extremely rare and poorly understood kind of cancer. These tumors resemble a different class of tumors known as signet ring cell adenocarcinomas, which exhibit mucin inside their cells and nearly always have a poorer prognosis than ordinary adenocarcinomas.2 Our case had the chief complaint of blood coming out of the urethra. Other symptoms that are found in urethral cancer include dysuria, dyspareunia, haematuria, perineal pain, urine retention, overflow incontinence, urethral mass, or a projecting meatal mass. It expands locally into the periurethral tissue, vagina, and vulva before spreading

Figure 1. Histopathology of the urethral lesion showing signet ring cell.
proximally to the bladder neck. Patients may present with a wide range of symptoms, the majority of which include dysuria, urine frequency, and a palpable mass. These three symptoms account for 50% of all presenting symptoms. Because of the shorter length of the female urethra, local spread tends to be more destructive.

According to research by Neto et al., their patient was hospitalized with stranguria, urine retention, and sero-bloody discharge with stoppers of mucopurulent material in addition to discomfort and burning in the urethra. The patient received surgery therapy such as extended radical vulvectomy and urethrectomy, progressing with the use of indwelling urinary catheter, as well as two sessions of radiotherapy. In this case, partial urethrectomy seems adequate not only because of the clear ability to distinguish healthy tissue and tumor tissue, but also to maintain the integrity and function of the lower urinary tract. Another study from Satyanarayan et al. reported a case with mucinous urethral adenocarcinoma. They performed a wide local excision of the tumor and underwent radical cystectomy with ileal conduit. The patient had normal postoperative physical examination results and imaging studies. But during the cystectomy she had persistent microscopic invasive adenocarcinoma in the residual urethra. A recurrence could be avoided with early detection and surgical intervention.

**Figure 2.** Current clinical appearance. Written informed consent was obtained from the patient for the use and publication of this clinical image.

**Figure 3.** Pelvic MRI (no residual mass or lymphadenopathy).
One recent study from Baffigo et al. reported a patient with mucinous adenocarcinoma of the bladder. The patient underwent anterior pelvectomy. Six months after surgery, bilateral inguinal lymph node dissection was performed because of bilateral palpable masses and the patient also received external radiotherapy of the inguinal area. After 22-months the patient appears healthy.5

A more recent study from Ndiaye M et al. reported a patient with urethra adenocarcinoma underwent partial urethrectomy. After 16 month follow up, the patient was continent without local and metastatic recurrence.6

From this case, with long term follow up we assessed response of urethral adenocarcinoma treated with radiotherapy. A limitation of this study is that the primary cause of the adenocarcinoma is still uncertain. In this case, we learned that performing partial urethrectomy is possible and able to maintain continence. Recurrence had appeared in this patient, but after radiotherapy, the patient currently remains clear from tumor.

**Conclusion**

Mucinous adenocarcinoma of female urethra is rare and there is no consensus on the optimal therapy due to the scarcity of cases. Early surgical treatment with or without adjuvant radiotherapy appears to be the best option in cases of small, organ-confined disease. Partial urethrectomy can be performed in patients with mucinous urethral adenocarcinoma, which can prevent the use of permanent urinary catheters and further improve the patient’s quality of life.

**Consent**

Written informed consent for publication of their clinical details and clinical images was obtained from the patient.

**Data availability**

All data underlying the results are available as part of the article and no additional source data are required.

**References**


Open Peer Review

Current Peer Review Status: ?

Version 1

Reviewer Report 14 July 2023

https://doi.org/10.5256/f1000research.145213.r185106

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This paper is well written in a good English. The explanation about the patient clinical condition may be missing some details, but enough for the reader to understand the big picture. Magnetic Resonance Imaging should have been performed in the first place instead of MRI, but it is understandable since the reader was not know at first that it was a urethral tumor. The author may suspect it was bladder tumor so I can understand. I would like the authors to provide the CT Scan and urethroscopy finding in this manuscript so the reader could relate with the author’s point of view. If the author could not provide those two due to some reasoning, at least the authors should elaborate those finding in a few sentences.

I would like to ask a question about the clinical reasoning on why the patient was not given chemotherapy. If the histopathological finding was adenocarcinoma, what is the consideration of the writer to not give the patient chemotherapy regimen for adenocarcinoma like Folfox or Folfiri regimen.

I think, the author should also emphasize that there is no fistulae when performing urethroscopy to exclude tumor infiltration from other organs.

That is my suggestion to improve this manuscript. Overall, this manuscript is good enough. However, there is a lot of room for improvement for this manuscript.

Is the background of the case’s history and progression described in sufficient detail? Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Partly

**Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?**
Yes

**Is the case presented with sufficient detail to be useful for other practitioners?**
Yes

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Urology, Oncology, Nanotechnology, Biomaterials, Tissue Engineering, Pediatric, Reconstructive Surgery, Stone Surgery

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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