Introduction

Physicians occasionally come across urethral lesions in women. A conclusive diagnosis is usually possible from a careful history, physical examination and use of adjunctive tests when necessary. 1 Few differential diagnosis are urethral caruncle, urethral prolapse, periurethral gland abscess, urethral diverticulum and malignancy. The hypoestrogenic state of the vaginal mucosa predisposes to urethral caruncle formation. Urethral carcinoma is usually encountered in the fifth to sixth decade of life. 2 Non-invasive squamous lesions are very rare with an uncertain clinical significance. We present a case of a premenopausal woman with urethral squamous papilloma having significant symptoms.

Case report

A 22 year old female presented with a history of abdominal distension, intermittent hematuria, occasional blood stained undergarments, dysuria, progressive obstructive symptoms such as straining to void, poor urinary flow and incomplete voiding for several months. She had autism with some difficulties in social interactions but could get along with her basic daily activities. Most of the history was provided by her mother. She had no issues with toilet training but had several episodes of urinary tract infection (UTI) in her childhood. She had never undergone any surgery before. She was unmarried and had no sexual history. There was no identifiable risk factor for urethral malignancy. Physical examination revealed pallor, lack of breast development and secondary sexual characters, tender palpable urinary bladder and an approximately 2 cm × 1.5 cm polypoidal erythematous mass arising just anterior to the vaginal introitus. (Fig. 1). Urethral meatus could not be separately delineated. The mass was friable, circumferential and bled on touch. She was catheterised with extreme difficulty under aseptic conditions with 6Fr Foley catheter through the centre of the mass thus confirming its circumferential origin from the urethra.

Serum chemistry revealed anaemia, mild leucocytosis and normal renal function test. Hormonal evaluation, including follicle stimulating hormone, luteinizing hormone and prolactin were within normal range. Thyroid stimulating hormone was raised. Urine analysis showed pyuria (8–10 cells/HPF) and microscopic hematuria. Peripheral smear revealed microcytic hypochromic red blood cells probably due to iron deficiency anaemia. Urine culture was positive for E. coli. Ultrasonography showed right sided hydrenephrosis and hydroureter with multiple bladder diverticula and thickened bladder wall with significant post void residual urine of approximately 250 cc. Uroflowmetry was not informative as her voided volume hardly exceeded 40 cc. No flow pattern could be generated. Pelvic floor muscle assessment was not done. She underwent Contrast enhanced CT scan of kidney, ureter and bladder which corroborated the ultrasonography findings except for mild left sided hydrenephrosis. Urinary bladder had multiple diverticula with contrast pooling even after voiding (Fig. 2).

She underwent excisional biopsy of the lesion under spinal anaesthesia. The mass extended a few millimetres into the urethra. It was completely excised with meatal reconstruction. Cystoscopy was done separately delineated. The mass was friable, circumferential and bled on touch. She was catheterised with extreme difficulty under aseptic conditions with 6Fr Foley catheter through the centre of the mass thus confirming its circumferential origin from the urethra.

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which showed normal proximal urethra with multiple diverticula in the bladder without any synchronous growth. The left ureteric orifice could not be visualised. Histopathology revealed urethral squamous papilloma (Fig. 3).

The patient was catheterised with 14 Fr Foley which was omitted the next morning. The patient voided well but she still had a feeling of incomplete evacuation with lower abdominal distension. She and her mother were explained and demonstrated the need for double voiding as well as clean intermittent self-catheterisation (CISC) after every void to completely empty the bladder.

The patient was followed up 2 weeks after surgery and now is on a 3 monthly follow up. She was also prescribed 75 mcg of L-thyroxin by an endocrinologist. She is doing well with no fresh episodes of UTI. She was very compliant with CISC even on her latest follow up visit. She has normal serum biochemical parameters. Repeat sonography did not show any upper tract dilatation but had multiple diverticula in the bladder with post void residual urine of approximately 90 cc. We have decided to keep her on follow up and treat her conservatively. We may address the diverticula, if she develops recurrent urinary tract infection with features of renal insufficiency in future.

Discussion

Urethral squamous papilloma is usually reported in women in their 40s–70s. There are very few documented cases in the literature.

Irritative voiding symptoms are the commonest presentation. Risk factors are supposedly similar as in urothelial carcinoma, including smoking and aromatic amines.

These lesions resemble low grade papillary urothelial carcinoma. Histopathological examination reveals squamous cells lining a fibrovascular core. The squamous cells may have koilocytotic changes or atypical features. In a study, urothelial squamous papilloma was found distinct from both verrucous carcinoma and condylomata acuminate, with no association with HPV infection. In addition, there was no mutation of p53 tumor suppressor gene as in verrucous carcinoma.

Urothelial carcinoma needs to be ruled out. It is best done by pathological analysis of excisional biopsy specimen by an experienced pathologist. HPV can be ruled out with p16 immuno staining. Recurrence is rare but data is limited.

The largest review was of 5 patients with squamous papilloma. Of these 5 patients, 2 were recurrence free after 15 months of follow up, 2 lost to follow up and 1 underwent cystectomy with urethrectomy after 21 months of follow up for urothelial carcinoma, but this patient was diagnosed with low grade urothelial cell carcinoma prior to the discovery of squamous cell papilloma.

This is just a case report so no recommendation can be made about the follow up protocol. However periodic assessment by urologist for recurrence of symptoms and imaging when needed is a reasonable follow up strategy. We cater to the need of a population which is
economically challenged with limited availability of health care services. This case may well be just a tip of an iceberg which gives us more of a responsibility to use all the urological armamentarium for the wellbeing of such patients.

Conclusion

Urethral squamous papilloma is a rare diagnosis with minimal malignant potential. It presents with both voiding and storage symptoms as well as with hematuria and renal insufficiency. It is primarily managed with excisional biopsy for therapeutic reasons as well as to rule out urothelial malignancy.

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Author contributions

Soumish Sengupta: Conceptualization, methodology, software, validation, formal analysis, investigation, resources, data curation, writing-original draft, visualization, project administration. Supriya Basu: data curation, supervision. Kadambari Ghosh: Conceptualization, writing-review and editing. Subhrajyoti Sengupta: data curation, writing-review and editing.

Declaration of competing interest

There are no conflicts of interest.

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