CASE REPORT

Vesical Leukoplakia: A rare entity

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Abstract:
Leukoplakia is an uncommon diagnosis. Patient usually presents with hematuria (visible and non-visible), dysuria, urgency and frequency. Definitive investigation is rigid cystoscopy. Abnormal bladder wall tissue should be resected and biopsies sent for histopathology to confirm the diagnosis. The condition is said to be pre-malignant it is usually advised to do annual cystoscopy with multiple biopsies is to look for any evidence of malignant transformation.

Case Report:
We present case report of a 64-year-old male presented to the urology clinic with a hematuria. Cystoscopy revealed a 2-cm white lesion on the right side of the urinary bladder. White plaques were also noted covering some areas of the bladder. Transurethral resection of the lesion and biopsy of the white plaques were performed. Pathological examination confirmed a diagnosis of keratinizing squamous metaplasia of the bladder with no evidence of malignancy.

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Introduction:
Vesical leukoplakia also known as Keratinizing squamous metaplasia (KSM) of the urinary bladder, is an abnormality of the urothelium in which the urothelium is transformed into a squamous cell epithelium beneath a layer of keratin. Mostly the patients present with non-specific urinary symptoms, including frequency, urgency, and other lower urinary tract symptoms (LUTS), macroscopic/microscopic hematuria, and genitourinary discomfort or pain. In clinical practice, urinary symptoms are the major concerns that prompt patients to seek treatment.[1]

Though the true etiology remains obscure, most writers strongly favor chronic infection or chronic irritation as the most likely cause. Another theory is that vitamin-A deficiency may be a factor in causation, and a number of cases have been associated with a positive Wassermann reaction. The bladder appears to be the most common site of leukoplakia in the urinary tract, and the renal pelvis to be less often affected.[2]

Diagnosis is usually made by cystoscopy with biopsies. Characteristic cystoscopy findings are white plaques in urinary bladder. Biopsy will confirm the diagnosis. Treatment includes transurethral resection and follow up with cystoscopy. Here we report one such rare case.

Case Presentation:
A 64-years old male patient who has no known co morbids presented to urology outpatient department with two months history of on and off lower urinary tract symptoms (LUTS) and hematuria. He was also complaining of passing white plaques in urine and sensation of foreign body while upon micturition. Physical and genitourinary examination was normal. Urine routine report showed the presence of red blood cells and epithelial cells. Ultrasound abdomen

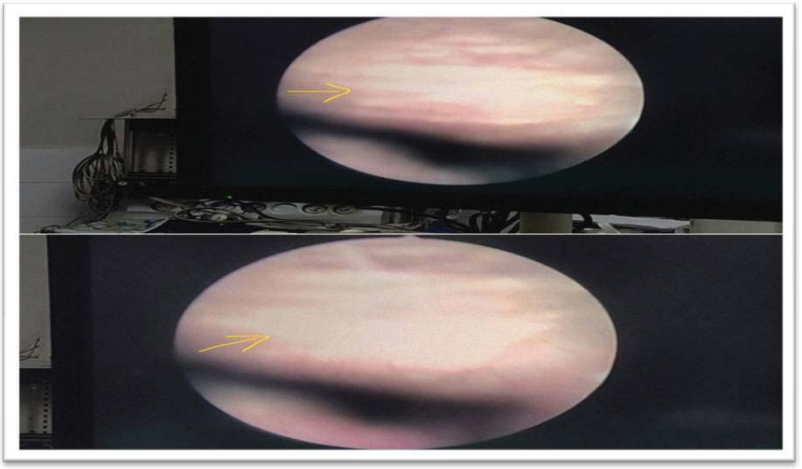

Figure 1: Cystoscopy images of the whitish plaques at right side of bladder wall, as pointed by yellow arrow

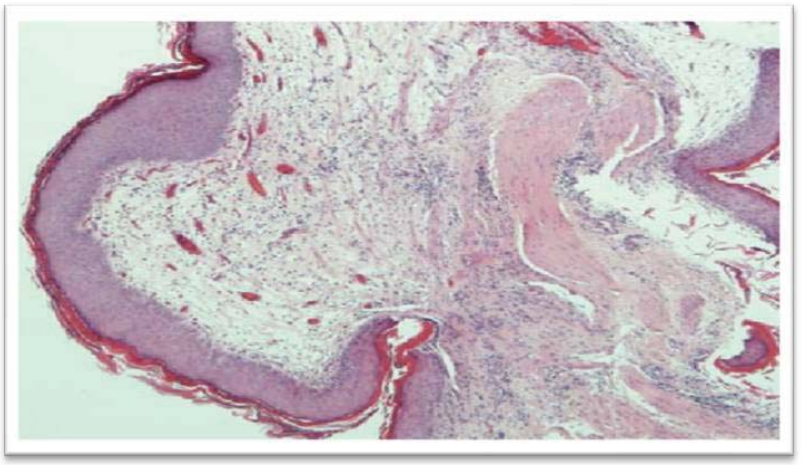

Figure 2: Bladder biopsy surfaced by keratinized squamous epithelium with edematous and mildly inflamed submucosa. (Picture taken from an article which is acknowledged with due reference (11))

and pelvis showed 45 grams prostate. He was admitted for diagnostic urethro-cystoscopy. After a detailed pre procedure workup and informed consent he underwent urethro-cystoscopy under spinal anesthesia. The findings were a 2-cm white lesion on the right side of the urinary bladder with white plaques were noted covering some areas of the bladder (Figure 1). Transurethral resection of the lesion and biopsy of the white plaques were performed. Pathological examination confirmed a diagnosis of leukoplakia bladder with no evidence of malignancy (Figure 2). Post-procedure recovery of the patient was uneventful and he was sent home on oral medications with follow up advised after 3 months for review.

Discussion:

Vesical leukoplakia is a very rare condition. Extensive literature searches on PubMed and Google Scholar yielded very limited literature. Upto 42% of cases are known to develop cancer, making early diagnosis critical. Patients with symptoms of bladder irritation and shedding of flakes are characteristic.[3] In our case, the patient complained of passing white flakes in the urine and a foreign body sensation during urination.

Rokitansky first identified the pathological characteristics of keratinizing squamous metaplasia in 1862 as excessive squamous epithelium development with desquamation and epithelium piling up. He gave the lesion the name "cholesteatoma". McDonald gave the most accurate definition of keratinizing squamous metaplasia, referring to it as the cornification of non-cornifying membrane (as cited by Marion).[4]

Keratinizing squamous metaplasia is seen during cystoscopy as a hyperemic region with distinct or diffuse whitish grey or pearl-like plaques. Plaques that have been removed reveal an underlying surface that is papillary and readily bleeds on instrumentation. The ureteric orifices are typically unaffected by keratinization, which can damage any region of the bladder, including the prostatic urethra. The lesions reveals yeast cystitis, alkaline encrustation, malakoplakia, and amyloid, which may resemble keratinizing squamous metaplasia on gross examination, are excluded by biopsy confirmation.[5]

Following the diagnosis, it is customary to perform cystoscopy monitoring to check for any potential malignant transformation.[6] The transitional epithelium of the bladder may sporadically experience an epithelial transformation into keratinizing squamous metaplasia as a result of exposure to persistently irritating stimuli, according to the pathophysiology of leukoplakia. Infection and chronic inflammation are the two of these triggers. Patients with spinal cord injury (SCI), for example, who frequently utilize chronic indwelling catheters for bladder man-

agement are more likely to develop this condition.[7]

The co-existence of squamous metaplasia and bladder cancer, especially squamous cell carcinoma, has been known since the disease was described. The first case of bladder cancer that occurred in a patient during long-term follow-upwas reported by Holley and Mellingerin1961.[8]

Since then, many cases of metachronous cancer have been reported. Benson et al, reported a 21% incidence of bladder cancer in a series of cases with keratinized bladder metaplasia. Therefore, it is accepted as a pre-malignant lesion or as a heralding lesion for the presence of malignancy elsewhere in the bladder.[9]

Vitamin-A deficiency was widely known as a cause several years ago. Wolbach and Howefed observed vitamin-A deficient rats and observed the formation of keratinized epithelium in the respiratory, uterine, and urinary tracts. As noted by Wins Bury-White, this finding was reinforced when Wilson and Dubois described a child who died of vitamin-A deficiency and suffered from extensive keratosis involving the urinary tract.[10]

We followed the same approach in diagnosing and treating patients as described in the literature. The purpose of reporting this case is to add scientific information about this condition to the body of knowledge for future reference and to assist urologists/nephrologists and physicians in diagnosing such patients in a timely manner.

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Abdul Haseeb, mian idea, manuscript writing and finalizing.

Sulaiman Shah, data retrieval, introduction writing.

Muhammad Idress Khan, data retrieval, discussion writing.

Muhammad Tayyib, data retrieval, references writing.

Jamal Ahmad Shah, data retrieval, Introduction writing.

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