

CASE REPORTS

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Cystitis glandularis- rare cause of urinary bladder mass: case report and literature review

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Abstract

Background: Cystitis glandularis a rare benign proliferative disease of the urinary bladder mucosa is usually a microscopic finding and manifests rarely as a large visible macroscopic lesion of urinary bladder. Only few cases of cystitis glandularis presenting as urinary bladder mass in female patients have been reported in the literature. We are reporting a case of cystitis glandularis presenting as a urinary bladder mass in a very young female patient.

Case presentation.

Our patient, a young female 25 years of age presented in the Urology OPD with complaints of irritative lower urinary tract symptoms for two years and intermittent hematuria for one year. On evaluation, she was found to have a urinary bladder mass. Transurethral resection of her urinary bladder mass was done and on histopathological examination, it came out to be cystitis glandularis.

Conclusion: Clinical presentation of cystitis glandularis is variable. It may remain asymptomatic or could present as irritative lower urinary tract symptoms, hematuria, or mucus in the urine. It is normally a microscopic finding but could also present as an overt urinary bladder mass however this has been reported rarely in the literature.

Keywords: Cystitis glandularis, Hematuria, Urinary bladder mass, Young female

1 Background

Cystitis glandularis is a very rare benign proliferative disease of the urinary bladder mucosa. The commonly affected age group is fifth and sixth decades of life. It is usually a microscopic finding and manifests rarely as a large visible macroscopic lesion of urinary bladder. However, cases of cystitis glandularis presenting as urinary bladder mass have been reported in the literature, very few such cases have been reported in female patients [1, 2].

Semins MJ reported a case of 32 yrs old female patient who was being evaluated for miscarriage. During her workup, a urinary bladder mass was found on pelvic ultrasound. Its histopathological analysis was suggestive of cystitis glandularis [3].

Shigehara et al. have reported a case of cystitis glandularis forming a tumorous lesion with blueberry spots in a 49 yrs old female patient [4].

Harrison S et al. also reported a case of 49 yrs old female patient who presented with nocturia, dysuria, intermittent hematuria and sensation of residual urine. On evaluation, she was found to have urinary bladder mass and on pathological examination it came out to be cystitis glandularis [2].

We present a case of young female of 25 yrs in age, who presented with complaints of irritative lower urinary tract symptoms and intermittent hematuria. Her USG abdomen, CT Urography, and cystoscopic examination revealed a urinary bladder mass. A complete transurethral resection was done. Findings of histopathological analysis were consistent for cystitis glandularis. Our case report will add in the literature a young female patient with cystitis glandularis forming a urinary bladder mass.

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2 Case presentation

We present a case of young married female, 25 years of age who presented in the Urology OPD with complaints of irritative lower urinary tract symptoms for two years and intermittent hematuria for one year. Her lower urinary tract symptoms included increased urinary frequency, urgency, and nocturia. Her hematuria was gross,

intermittent, total, and sometimes associated with passage of clots. There was no history of fever and burning micturition. There was no history of obstructive lower urinary tract symptoms and acute urinary retention, any chronic illness like hypertension, diabetes mellitus, tuberculosis, coronary artery disease, etc., any other major complaints, any urological intervention or surgery in the past, and any kind of addiction. Her general and systemic examination were apparently normal and could not add any positive finding in her clinical case. Her haemogram, kidney function test, liver function test, and coagulation profile were within normal range except for slightly decreased hemoglobin. Her urine microscopic examination was positive for hematuria, however, her urine culture was sterile and urine cytology was negative for any malignant cells. Her USG whole abdomen was suggestive of a well-circumscribed soft tissue lesion in the urinary bladder near left vesicoureteric junction. Her CT urography was suggestive of a polypoid mass of size approximately 3.2×2.9 cm arising from inferoposterior and left lateral surface of urinary bladder and extending into bladder lumen (Fig. 1). Her cystoscopic examination revealed a single polypoid mass of size approximately 3×3 cm arising from inferoposterior and left lateral wall (Fig. 2). A complete transurethral resection of the mass was performed. Histopathological examination revealed



Fig. 1 CT urography suggestive of a polypoid mass of size approximately 3.2×2.9 cm arising from inferoposterior and left lateral surface of urinary bladder and extending into bladder lumen

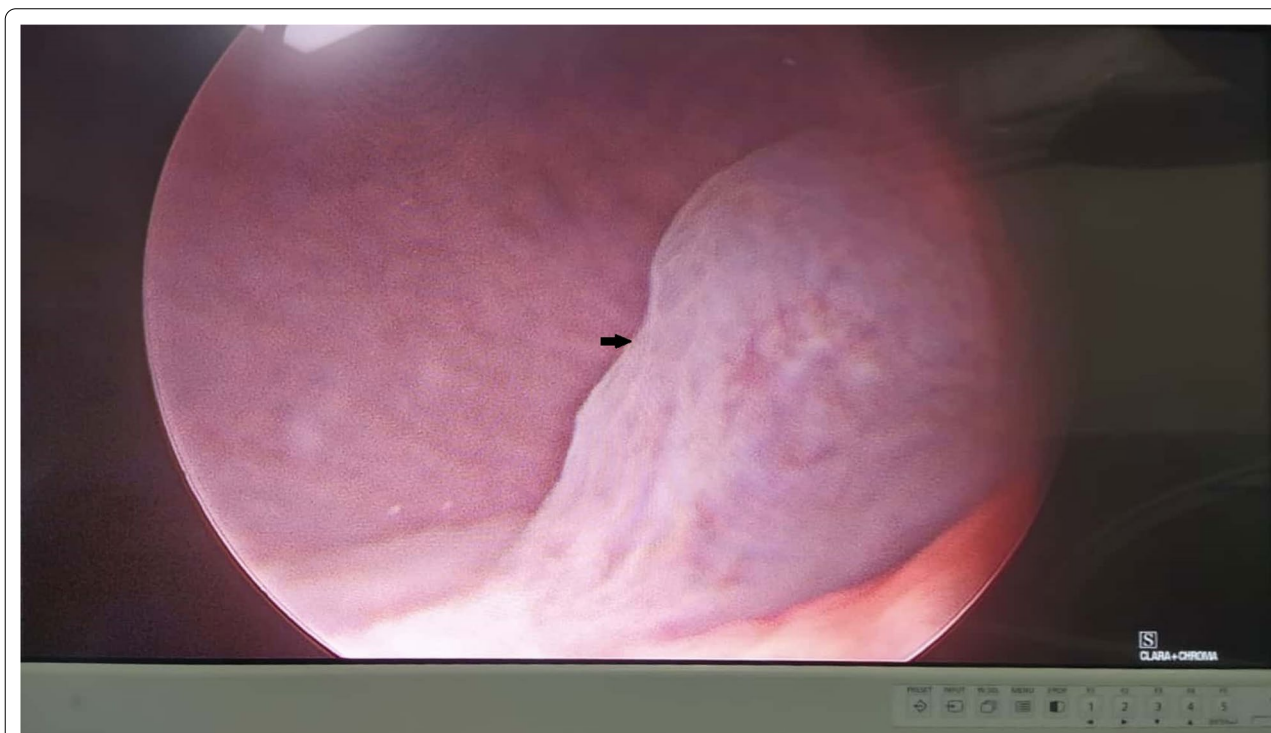


Fig. 2 Cystoscopic examination revealing a single polypoid mass of size approximately 3×3 cm arising from inferoposterior and left lateral wall. This is a close view showing cystoscopic appearance of polypoidal mass

gland-like lumens with columnar cells lining and surrounding chronic inflammatory cells infiltrate in the lamina propria (Fig. 3). These histopathological findings were suggestive of cystitis glandularis. There was no evidence of von Brunn nests, significant atypia, stromal reaction, muscular invasion, or any other evidence of malignancy on histopathological examination. The patient is in follow-up since her surgery almost one year back.

3 Discussion

Cystitis glandularis is a rare proliferative disorder of the mucus-producing glands within the mucosa and submucosa of urinary bladder epithelium [5]. It is commonly located on the lateral bladder wall, trigone, and the bladder neck [6]. It was first described by Morgagni in 1761 [7] and since then has been a topic of interest.

Different etiologies regarding its origin exist in the literature however it has been a matter of debate. One hypothesis believes it to be a result of abnormal embryologic development. As per this theory bladder, embryonic rests originate from either urachus or intestinal epithelium and get displaced during separation of the rectum from the urogenital sinus. However embryologic theory of origin was not accepted by all as cystitis glandularis also occurs in ureter and renal pelvis and currently this hypothesis has been abandoned. The second predominant hypothesis believes it to be a metaplastic change in the vesical mucosa due to inflammation and irritation. It leads to formation of Von Brunn nests along with degeneration of central cells and metaplasia of the surrounding cells to a columnar phenotype, which is consistent with cystitis glandularis. Many factors such as bladder exstrophy, pelvic lipohypertrophy, recurrent urinary tract infection, inflammation caused by urolithiasis, outflow

obstruction and indwelling urethral or suprapubic catheter are considered to be risk factors for development of cystitis glandularis. Different other suggested etiological factors include avitaminosis, allergy, toxic products of metabolism, hormone imbalance, and specific carcinogens [3, 4, 7–10].

Clinical presentation of cystitis glandularis is variable. It could remain asymptomatic or could present as irritative lower urinary tract symptoms including dysuria, increased frequency, and urgency. It could also present as hematuria or mucus in the urine. It is normally a microscopic finding and an overt urinary bladder mass is extremely rare in the literature [1, 5, 11]. Its diagnosis is based on histological findings, however nowadays the role of CT and diffusion-weighted MRI is also being explored [12].

Natural history of cystitis glandularis regarding its premalignant potential is also a controversial topic. Shaw et al. in 1958 described a case of cystitis glandularis which gradually converted into adenocarcinoma [13]. Edwards et al. also documented the progression of cystitis glandularis to adenocarcinoma [14]. However, Davies et al. found no clinical evidence to label it a premalignant condition [7]. Similarly, Smith et al. in their study reported that Cystitis glandularis can be identified coincidentally with bladder carcinoma relatively frequently, although the long-term outcomes of their patient population have not supported the role of this condition as preneoplastic [15]. The results of Yi et al. also did not supported the premalignant potential of cystitis glandularis in their study, however, follow-up period in this study ranged from 0.5 years to 17 years [16].

Apart from management of risk factors of cystitis glandularis (inflammation and obstruction) and complete transurethral resection of the urinary bladder lesion, different other treatment modalities have also been reported in the literature. Yuksel OH et al. have reported the use of oral steroid for the treatment of intractable cystitis glandularis [17]. Takizawa et al. reported the use of celecoxib (a non-steroidal anti-inflammatory COX-2 selective inhibitor) for treatment of refractory cystitis glandularis [18]. Stillwell et al. reported use of whole bladder neodymium:YAG laser therapy for a case of severe cystitis glandularis of the entire bladder [19]. Black et al. reported a case of rapidly recurring cystitis glandularis who developed a low compliance small-capacity bladder and bilateral ureteral obstruction. They performed total cystoprostatectomy with cavernosal nerve-sparing and ileal neobladder construction [20].

Endoscopic resection is usually sufficient for symptomatic relief of patients, however, for severe and recurrent cases, extensive surgical management may be needed. After transurethral resection annual surveillance by

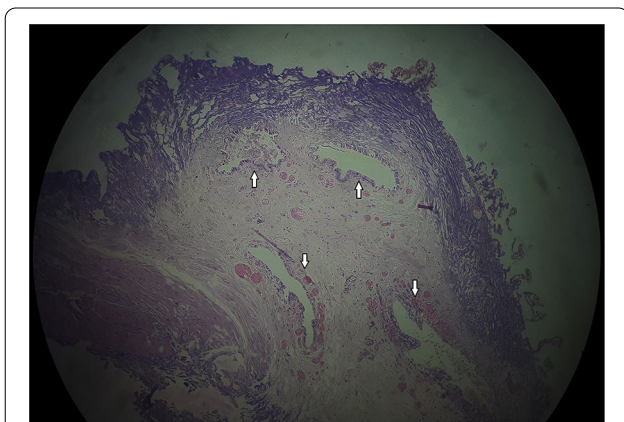


Fig. 3 Histopathological examination revealing gland-like lumens with columnar cells lining and surrounding chronic inflammatory cells infiltrate in the lamina propria (H&E, 100X)

cystoscopy with bladder biopsies is recommended, owing to risk of progression from cystitis glandularis to adenocarcinoma. Imaging and urinary cytology is also advocated [2].

Hu J et al. in their study revealed that the urinary infection, long-term indwelling catheter usage, urinary calculus, squamous metaplasia, and atypical hyperplasia were independent risk factors for CG recurrence [21].

4 Conclusions

Cystitis Glandularis is a rare proliferative condition of urinary bladder and usually presents as a microscopic finding. It is very rare to find a large visible macroscopic lesion of urinary bladder due to cystitis glandularis. Many more studies and work are required on this topic to solve different associated controversies and debate.

Abbreviations

OPD: Out Patient Department; USG: Ultrasonography; CT: Computed tomography; MRI: Magnetic resonance imaging; COX-2: Cyclooxygenase 2; YAG: Yttrium aluminium garnet; H&E: Hematoxylin and Eosin.

Acknowledgments

We are thankful to radiologists for their important role in the workup of our patient.

Authors' contributions

A.A. and N.R. made the clinical decision of the case and were having leading role in operative procedure and have helped to draft the manuscript. R.K.T. and M.Z.I. have done the research work related to topic and participated in manuscript writing. B.K. was responsible for histopathological diagnosis, and provision of the pathological microphotographs. Q.K. was involved in describing pathological findings and manuscript writing. All authors have read and approved the manuscript.

Funding

No source of funds.

Availability of data and materials

Not applicable

Declarations

Ethics approval and consent to participate

Not Applicable.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests

The authors declare that they have no competing interests.

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Received: 31 July 2021 Accepted: 13 March 2022

Published online: 22 March 2022

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