Radiology Case Reports 19 (2024) 2357–2361

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Cystitis glandularis mimicking a bladder tumor: A rare case report ☆,☆☆

Imad Bougrine, MD, Kaoutar Imrani, PhD, Amine Naggar, MD, Hamza Retal, MD, Nabil Moatassim Billah, PhD, Ittimade Nassar, PhD*

Radiology department, Ibn Sina University Hospital, Mohammed V University, Rabat Morocco

ARTICLE INFO

Article history:
Received 30 November 2023
Revised 27 February 2024
Accepted 29 February 2024

Keywords:
Cystitis glandularis
Pseudotumoral
Imaging

ABSTRACT

Cystitis glandularis is a rare benign condition, occurring in only 1% of the general population, and it tends to affect males more frequently. This pathology is characterized by reactive metaplasia of the bladder's urothelium, which results from chronic and recurrent irritation of the bladder wall. Symptoms are nonspecific and primarily marked by an irritative urinary syndrome accompanied by hematuria. We present a case of a young male patient with cystitis glandularis, discovered as a result of bilateral uretero-hydronephrosis detected during an ultrasound examination subsequent to an irritative urinary syndrome. The diagnosis was initially suggested by a CT scan; it was ultimately histologically confirmed following the endoscopic resection of the tumor.

Introduction

Cystitis glandularis (CG) is a benign condition characterized by reactive metaplasia of the bladder urothelium [1]. It is a rare condition, predominantly affecting males, and is caused by chronic and recurrent irritation of the bladder [2]. The clinical incidence of the disease is estimated to be around 1% of the general population [3,4]. It often goes underdiagnosed, and its underlying pathophysiological mechanisms remain poorly understood. Symptoms are nonspecific, dominated by an irritative urinay syndrome and hematuria [5].

Case report

We present a case of a 28-year-old male patient with no notable personal or family medical history, who consulted for a persistent obstructive syndrome of the lower urinary tract progressing over 4 months and consisting of increased urinary frequency, recurrent hematuria, weak urine stream, and straining. Physical examination revealed pain in the hypogastric region, a good general condition, and no fever. The biological workup showed preserved renal function, and the cytobacteriological examination of urine revealed leuko-

☆ Acknowledgments: None.
☆☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.
* Corresponding author.
E-mail address: imad_bougrine@outlook.fr (I. Bougrine).
https://doi.org/10.1016/j.radcr.2024.02.114

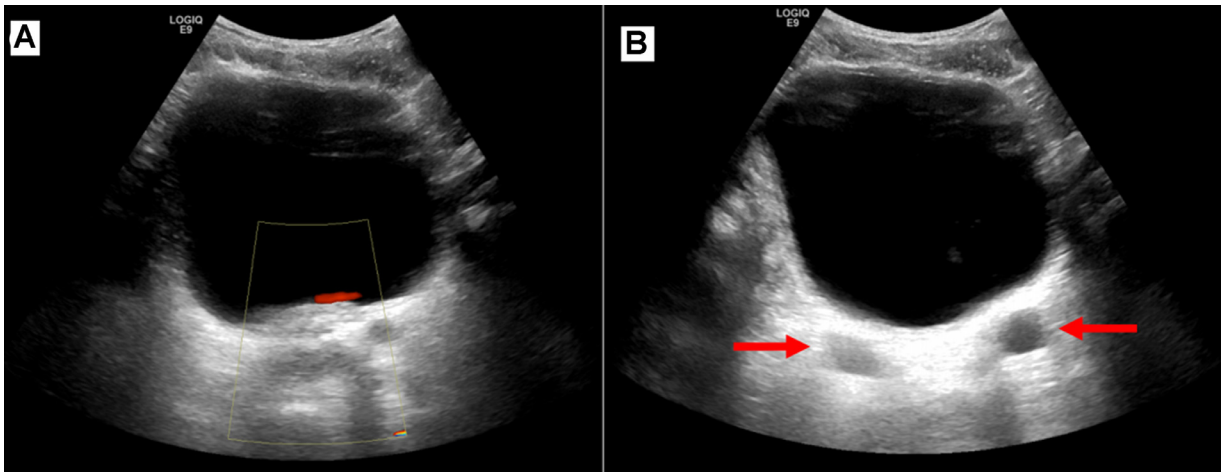

Fig. 1 – Ultrasound in axial view: vascularized posterior bladder wall thickening on color Doppler which infiltrates the ureteral meatus (A) with bilateral upstream uretehydronephrosis (B, arrow).

cyturia without bacterial isolation; no other biological abnormalities were found. A reno-vesico-prostatic ultrasound was performed, showing regular thickening of the posterior vesical wall, which appeared vascularized on color Doppler. There was infiltration of the ureteral orifices causing bilateral uretero-hydronephrosis (Fig. 1), with a normal renal cortical index (right renal pelvis = 25 mm, left renal pelvis = 30 mm); the prostate was of normal volume.

A uroscan was subsequently performed, revealing a posterior thickening of the bladder wall protruding into the bladder lumen, with postcontrast enhancement. This thickening infiltrated the ureteral orifices bilaterally but did not extend to the peri-vesical fat. Additionally, moderate bilateral uretero-hydronephrosis with tortuous ureters was observed (right ureter = 17 mm, left ureter = 19 mm), along with delayed secretion and renal excretion (Fig. 2). A cystoscopy was performed, showing an ulcerated solid vegetative lesion on the posterior bladder wall. A complete endoscopic resection was conducted, and histological examination confirmed the diagnosis of CG by revealing abundant urothelial Von Brunn nests (Fig. 3). Antibiotic therapy was prescribed for 2 weeks, resulting in improvement of clinical signs.

Discussion

CG is a proliferative disorder of the bladder mucosa, arising from reactive inflammatory processes in the context of chronic bladder irritation. It is frequently identified on biopsies or cystectomy specimens, with a preferential localization at the trigone. Despite numerous studies, the etiopathogenesis of CG remains poorly understood. Several theories, including the embryonic theory and the metaplastic theory, have been proposed to explain its development:

1. *The embryonic theory:* intestinal germ cells abnormally migrate towards the bladder during its separation from the rectum at the fifth week of embryonic development, leading to CG [6].
2. *The metaplastic theory:* the transitional epithelium undergoes metaplastic transformation in response to irritating agents such as lithiasis, prolonged urinary stasis, urinary infections, or tumors [7].

CG is associated with pelvic lipomatosis and bladder dystrophy in 75% of cases. These conditions serve as disease-promoting factors and pose a risk for malignant transformation into adenocarcinoma [8]. Other etiologies described in the literature include radiotherapy and chemotherapy [9]. Additionally, certain research suggests the involvement of an immune mechanism. Abnormal presence of IgA on the surface of transitional cells of the bladder has been observed in cases of CG, whereas normally IgA should be localized inside the cytoplasm of normal bladder cells [10].

Regarding clinical expression, it is often nonspecific; most cases are asymptomatic, although individuals may occasionally exhibit signs of irritability such as increased frequency, dysuria, or urinary urgency. However, hematuria remains the most common revealing sign [11]. In advanced stages of the disease, there is intramural compression of the distal ureteral segments, leading to the development of upstream uretero-hydronephrosis. This complication can be further complicated by late chronic renal failure [12]. Radiologically, the CT scan is the best imaging modality for studying the urinary system and for etiological investigation. It can show lithiasis and pelvic lipomatosis, and often exhibits a simulated resemblance to vesical carcinoma through tissue thickening and infiltration of the bladder's base [13]. Ultrasound often reveals focal polypoid wall thickenings in the trigone region [14]. It serves as an examination for orientation and evaluation. Magnetic resonance imaging (MRI) is generally not recommended for diagnosing due to the absence of specific differentiation signs from malignant bladder lesions [15], it shows polyploid wall thickening with low signal intensity on T1 and T2 weighted sequences, accompanied by a central branching high-signal-intensity pattern in T2 which en-

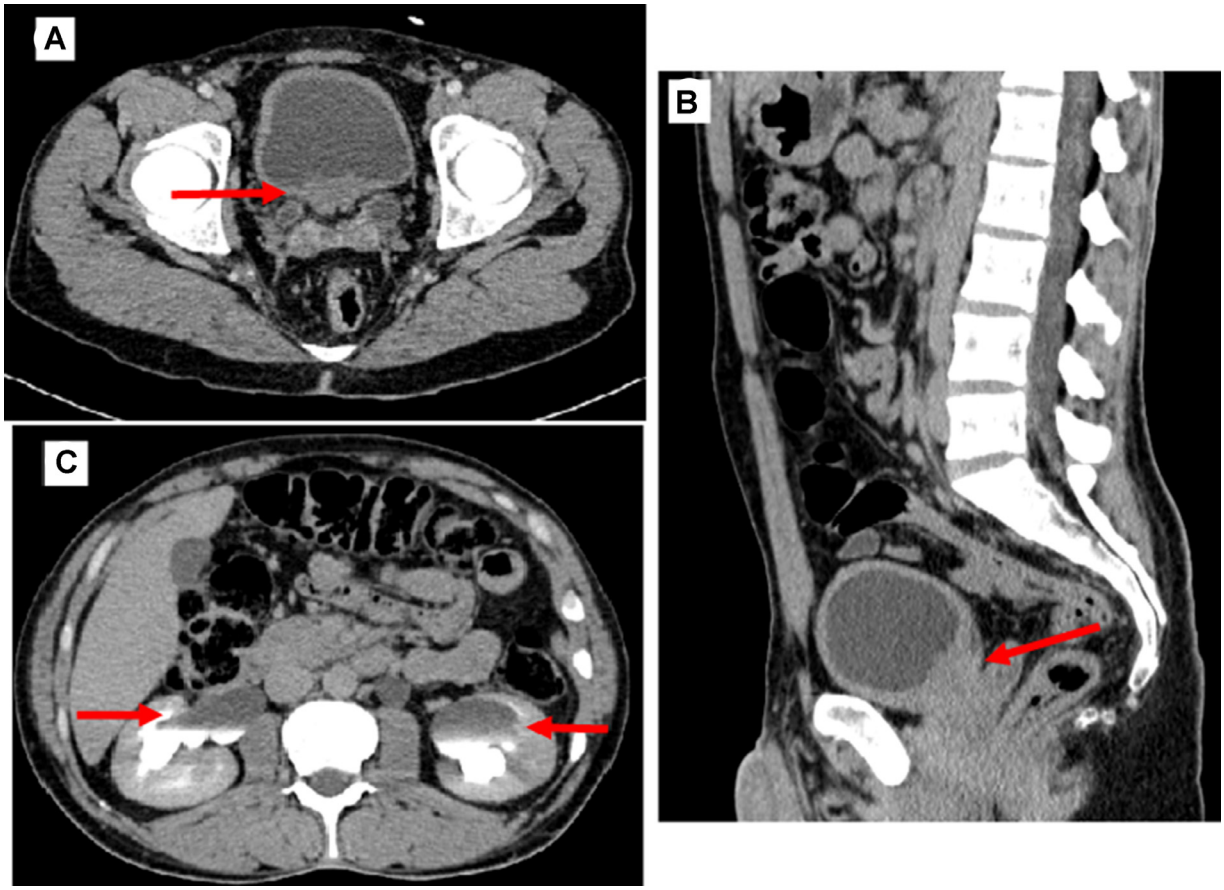

Fig. 2 – Computed tomography (CT) images showing posterior bladder wall thickening infiltrating the ureteral orifices (A, B) (arrow) with upstream ureterohydronephrosis and delayed secretion and renal excretion (C, arrow).

hances after injection of contrast product, corresponding to vascular ramifications [16]. Cystoscopy enables visualization of the lesion, its macroscopic appearance, its precise location, and facilitates the collection of samples for diagnostic confirmation [17].

Histologically, initially, the proliferation of urothelial cells up to the chorion leads to the formation of "islets of Von Brunn" which are urothelial cells presenting a lumen lined with secreting cells. they give rise to the formation of true glandular structures, which can take on a pseudo-colonic appearance with the presence of goblet cells. In some cases, this lumen expands to form cysts containing colloids [12]. Immunohistochemistry highlights characteristics not visible in simple histology. Chromogranin immunostaining shows the presence of spindle-shaped endocrine cells in the Von Brünn islets between the epithelial cells [18], which contain intracytoplasmic neuroendocrine granules positive for Grimelius silver staining.

The differential diagnosis between CG and bladder cancer holds paramount clinical significance in medical practice, the connection remains inconclusive, leading to numerous areas of disagreement [19]. As outlined by some doctors, there exists a link between CG and bladder cancer, suggesting that CG could be viewed as a precancerous lesion within the bladder. On the contrary, some argue that there is no apparent association [19]. Bladder cancer is commonly observed among elderly males. Most patients typically present with painless gross hematuria, often associated with inflammation of the urinary tract. Approximately 60% of bladder malignancies originate in the posterior wall, while 20% manifest in the trigone. Although CG can develop at any age, it is predominantly found in middle-aged women, frequently localized in the trigone and neck of the bladder. Chronic irritation of the urinary tract stands out as the primary clinical symptom [20,21]. Imaging examinations of bladder cancer lesions reveal an irregular surface with evidence of liquefactive necrosis and spot-like calcifications. The observed density is inconsistent and undergoes significant enhancement during contrast-enhanced scanning. Bladder cancer exhibits a higher CT value compared to CG. There is potential for bladder cancer to spread to the pelvic lymph nodes and infiltrate the muscular and adventitial layers of the bladder. In contrast, CG lesions present with a smoother appearance than those of cancer and display uniform interior density. Although density enhancement can occur during contrast-enhanced scanning, the effect is subtle. These lesions primarily originate from the submucosal layer without infiltrating the muscular layer or adventitia [22].

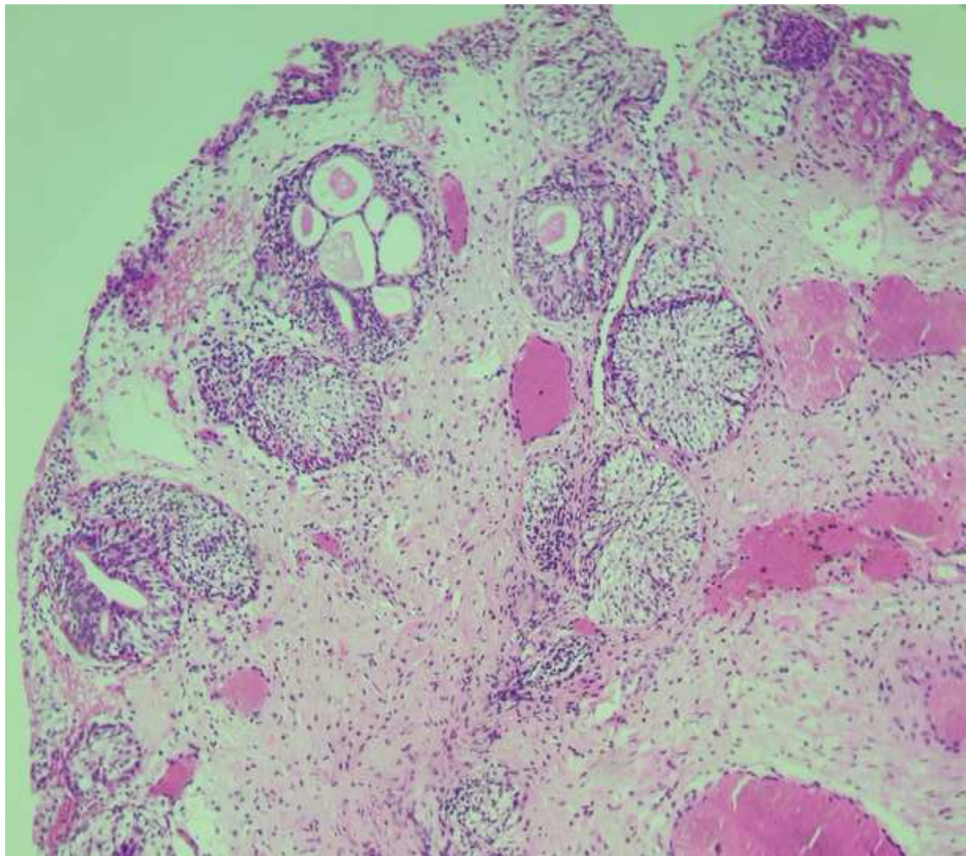

Fig. 3 – Von Brunn urothelial nests presenting glandular aspects, with cuboidal or cylindrical cells lacking atypia or mitosis, without sign of invasion.

Conclusion

Cystitis glandularis is a rare condition that affects the bladder wall and can cause an irritative or even obstructive urinary syndrome. The diagnosis may be made in a young nonsmoking individual with no family history of bladder cancer, but definitive diagnosis relies on histological examination.

Ethical Approval

No ethical approval is required for de-identified single case reports based on our institutional policies.

Patient consent

Written informed consent was obtained from the patient.

REFERENCES

[1] Jacobs LB, Brooks JD, Epstein JI. Differentiation of colonic meta-plasia from adenocarcinoma of the urinary bladder. Hum Pathol 1997;28:1152–7. doi:10.1016/S0046-8177(97)90253-7.

[2] Mast P, Casselman J. La cystite glandulaire: cause rare de masse vésicale. Cas clinique et revue de la littérature. Acta Urol Belg 1994;62:71–6.

[3] Bryan RT, Nicholls JH, Harrison RF, Jankowskid JA, Wallace DM. The role of beta-catenin signaling in the malignant potential of cystitis glandularis. J. Urology 2003;170:1892–6.

[4] Cabanne F, Pages A, Billeret C, Opperman A, Carbillet JP. Métaplasie glandulaire de la muqueuse urothéliale. Pathol Génitale Masc, Ed. Masson 1993:300–1.

[5] Granados EA, Algaba F, Vicente Rodriguez J. Cystitis glandularis. Arch Esp Urol 1999;52:119–22.

[6] Billerey C, Sibony M. Anatomie pathologique des tumeurs superficielles de la vessie. Progrès En Urol 2001;11(5):805–863.

[7] Borda A, Decaussin Petrucci M, Berger Nicole. Lésions bénignes diverses de la vessie et de la voie excréto urinaire. Ann Pathol. Elsevier Masson 2004;24:18–30.

[8] Fariña LA. Re: pelvic lipomatosis associated with cystitis glandularis and adenocarcinoma of the bladder. J Urol 1992;147(5):1380.

[9] Chan TY, Epstein JI. Radiation or chemotherapy cystitis with pseudocarcinomatous features. J. Urol 2004;28:909–13.

[10] Uehling DT. Secretory immunoglobulin: a excretion in cystitis cystica. Urol Volume 1973;1(4):305–6.

[11] Davies G, Osborn DE, Castro EJ. Pelvic lipomatosis with associated cystitis glandularis. Urology 1978;11(5):494.

[12] Darouichi M. Glandular and cystic bladder cystitis: case report and review of the literature. J Dig Disord Diagn 2020;2(1):17–23.

[13] Robert H, Dann PH, Arger E, Horatio T. Benign proliferation processes presenting as mass lesions in the urinary bladder. Am J *Roentgenol* 1972;116(4):822–9.
[14] Wong-You–Cheong JJ, Woodward PJ, Manning MA, Davis CJ. Inflammatory and non-neoplastic bladder masses: radiologic-pathologic correlation. Radiographics 2006;26(6):1847–68. doi:10.1148/rg.266065126.
[15] Rocher M, Claudon L, Cluzel C, Roy G. Imagerie des tumeurs rares et des pseudotumeurs de la vessie. J Radiol 2009;90(10):1415.
[16] Kim SH, Yang DM, Kim NR. Polypoid and papillary cystitis mimicking a large transitional carcinoma in a patient without a history of catheterization: computed tomography and magnetic resonance findings. J Comput Assist Tomogr 2004;28:485–7.
[17] Wong-You–Cheong JJ, Woodward PJ, Manning MA. Inflammatory and nonneoplastic bladder masses: radiologic-pathologic correlation1. Radiographics 2006;26(6):1847–68.
[18] Benchakroun A, Zannoud M, Nouini Y, Bernoussi Z, Kamouni M, et al. Métaplasie colique pseudo tumorale de la muqueuse vésicale. Prog Urol 2002;12:325–8.
[19] Li Z, Liu P, Zu X, Fan B. Progression of research into the diagnosis and treatment of cystitis glandularis: a narrative review. J Xiangya Med 2022;7:24.
[20] Filipas D, Fichtner J, Fisch M, Hohenfellner R. Pseudotumoral cystitis cystica of the urethra in a boy. Br J Urol 1997;79:656–7.
[21] Masuda M, Kitami K, Tïba K, Kumagai H. A case of cystitis cystica and glandularis suspected of submucosal tumor of urinary bladder. Hinyokika Kiyo 1989;35:505–8.
[22] Hua H, Gao Y, Lin J, Hou F, Wang JW, Zhang Y, et al. Quantitative analysis of enhanced computed tomography in differentiating cystitis glandularis and bladder cancer. Biomed Res Int 2020;2020:4930621.