



Case Report

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Intestinale Florid Glandular Cystitis in Centre Medicochirurgical De Kinindo Bujumbura

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Abstract

Background: Glandular cystitis is a metaplastic lesion of the bladder urothelium primarily affecting men. There are two types: typical glandular cystitis and intestinal glandular cystitis. Therefore, the definitive diagnosis relies on histopathology. The controversial theory surrounds the malignant degeneration of these lesions. Here, we present a case of florid glandulo-cystic cystitis of intestinal origin. Clinic observation: A 20-year-old man presented to the clinic with intermittent total hematuria associated with obstructive and irritative symptoms that had been ongoing for more than a year. In Histology examen, there was an absence of malignant tumor proliferation and signs indicative of typical inflammation: Florid glandulo-cystic cystitis of intestinal type. Conclusion: Intestinal glandular cystitis is a rare condition affecting primarily men and involving the bladder mucosa. Its symptoms are nonspecific and often go undiagnosed. Transurethral bladder resection remains the treatment of choice, but other therapeutic modalities, proven effective, should not be dismissed. The issue of malignant degeneration remains a controversial topic for debate, but the course of the disease is primarily marked by recurrences, necessitating cystoscopic monitoring.

Keywords: Intestinale, Florid glandular, Cysitis, Bujumbura.

INTRODUCTION

Glandular cystitis is a metaplastic lesion of the bladder urothelium primarily affecting men ^[1, 2]. There are two types: typical glandular cystitis and intestinal glandular cystitis. Glandular metaplasia of the intestinal type, exhibiting the appearance of complete intestinal metaplasia with mucosecretion and the absence of persistent urothelial cells, can be observed in response to chronic irritative or inflammatory processes such as neurogenic bladders, bladder exstrophy, prolonged catheterization, or a history of calculi ^[3]. The symptoms of intestinal-type glandular cystitis are often atypical, posing a challenge in the differential diagnosis with malignant bladder tumors. Therefore, the definitive diagnosis relies on histopathology. The controversial theory surrounds the malignant degeneration of these lesions.

Here, we present a case of florid glandulo-cystic cystitis of intestinal origin.

CASE REPORT

A 20-year-old man presented to the clinic with intermittent total hematuria associated with obstructive and irritative symptoms that had been ongoing for more than a year. Urological history and physical examination were otherwise unremarkable. Laboratory tests were within normal ranges. Abdomino-pelvic ultrasound revealed irregularity of the left lateral-cervical bladder mucosa and a significant post-void residual volume. Prostatic volume was estimated at 33g, and both kidneys were of normal size.

During urethro-cystoscopy, the urethra appeared normal, and a round, whitish tumor was observed in the trigone, protruding into the bladder neck. The bladder mucosa exhibited a vibrant appearance reminiscent of a schistosomiasis-infected bladder. Endoscopic resection of the tumor was performed, with hemostasis and the placement of a double-current urinary catheter. The postoperative course was uneventful, and the catheter was removed the following day.

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In Histology examen, a total of 2g tissue samples were completely included. A normal surface urothelium was observed. The superficial and deep chorion contained numerous Von Brunn nests undergoing central cavitation filled with mucin. These cavities were lined by a single-layered muciparous cylindrical epithelium of intestinal type. There was an absence of malignant tumor proliferation and signs indicative of typical inflammation: Florid glandulo-cystic cystitis of intestinal type.

Outcome: On the third postoperative day, the urine was clear, and there was improvement in lower urinary tract symptoms. The patient was scheduled for a cystoscopic follow-up appointment within six months after discharge.

DISCUSSION AND LITERATURE REVIEW

Glandular cystitis is a rare benign tumor of the bladder mucosa that originates from Von Brunn nests. There are two types: typical glandular cystitis (non-mucinous), which is more common, and intestinal glandular cystitis (mucinous), which is rarer.

Intestinal-type glandular cystitis exhibits the appearance of complete intestinal metaplasia with mucosecretion, without the persistence of urothelial cells [4]. In the majority of cases, this glandular metaplasia affects the bladder trigone, but it can also extend to the level of the ureter and renal pelvis. The pathophysiology is not well understood. However, chronic irritative and inflammatory processes of the bladder mucosa are the most incriminated risk factors, including neurogenic bladders, bladder exstrophy, pelvic lipomatosis, prolonged catheterization, or a history of calculi [4-6].

Other factors such as avitaminosis, allergies, hormonal imbalance, specific carcinogens, and IgA-mediated immune mechanisms have been mentioned in the literature [7,8]. Another etiological hypothesis is based on embryonic rearrangements originating in the urogenital sinus, leading to the formation of true enteric glands, with or without mucous content in the bladder. Mucosal metaplasias can also result from embryonic remnants of the skin (squamous metaplasia) or a primitive kidney (nephrogenic adenomas) [9].

Coelho R.F. and colleagues reported a case of severe intestinal glandular cystitis in an HIV-immunocompromised patient. However, the exact correlation between immunosuppression and glandular cystitis was not specified [10]. In our case, a subvesical obstruction was the most strongly implicated contributing factor. The pre-neoplastic nature of glandular cystitis remains a contentious issue [11]. Xil et al. [3] monitored 89 patients over 105 months, reporting a case that progressed to adenocarcinoma in a patient with high-grade metaplasia. In a 2015 study by Gordetsky and Epstein [12] involving 19 cases of glandular cystitis with dysplasia, 8 patients later developed bladder adenocarcinoma. Corica et al. [6] followed 53 patients with intestinal glandular cystitis over 10 years, and none developed bladder adenocarcinoma. XIANLIN YI et al. [13] found no cases of bladder adenocarcinoma among 166 cases of glandular cystitis followed for 17 years post-treatment.

The incidence of glandular cystitis is 60 to 70% in autopsy cases and 0.1 to 1.9% in clinical settings. However, the true incidence is challenging to determine due to the often vague and nonspecific symptoms associated with this condition [3,5]. Intestinal glandular cystitis is typically found in patients in their 5th and 6th decades and more commonly affects men than women [3,14]. Nevertheless, cases in children have been reported, and individuals of all ages can be affected, ranging from 3 to 82 years [15]. In the literature, only four cases in women have been mentioned by Schoenberg [16], Shigehara [17], Sumba [18], and more recently by Ahmad et al. [19].

The symptoms of glandular cystitis are nonspecific. In the majority of cases, it is asymptomatic and often underdiagnosed. The milder form may be asymptomatic or present with signs of common cystitis. The

more pronounced pseudotumoral forms are symptomatic in one-third of cases. Two symptomatic patterns have been described in the literature: intermittent hematuria alone or associated with other urinary symptoms predominates in two-thirds of cases. On the other hand, irritative symptoms such as urgency, burning on micturition, diurnal and nocturnal polyuria can occur. Hematuria accompanied by mucosuria is also possible. Physical examination is often unremarkable [18-22]. In our context, hematuria was associated with obstructive and irritative symptoms, a combination also noted by other authors such as Sumba H. et al. [18].

The diagnosis of glandular cystitis is primarily based on histological results, but currently, the role of CT scans and diffusion-weighted MRI is under exploration [23]. There are several therapeutic modalities, ranging from conservative to more aggressive approaches.

Firstly, identifying, treating, and eradicating all factors that contribute to chronic bladder irritation constitute a crucial aspect of management. This involves appropriate antibiotic therapy to address urinary infections, promoting intermittent self-catheterization instead of indwelling catheters, or treating bladder calculi. Symptomatic patients may benefit from transurethral resection. These conservative measures are suitable for small-localized lesions, as was the case with our patient [24]. Yuksel OH et al. [25] reported the use of oral steroids for the treatment of recurrent glandular cystitis. Takizawa et al. [26] have mentioned the use of Celecoxib (a selective COX-2 inhibitor NSAID).

Endovesical instillations of hydrocortisone, DMSO, low molecular weight heparin, or antihistamines can be employed. However, these endovesical instillations appear to improve symptoms without necessarily reducing the size of the tumor [27]. The use of YAG laser has been described.

Surgical interventions of varying degrees are performed in cases of ureteral meatus invasion (bilateral ureterovesical reimplantation) or debilitating irritative phenomena (partial cystectomy, total cystectomy). Black et al. [28] reported a total cystoprostatectomy preserving the cavernous nerve with an ileal neobladder in a case of rapid recurrence that developed low compliance, small bladder capacity, and bilateral ureteral obstruction

Coelho R.F. et al. [10] described a case of severe glandular cystitis in an immunocompromised HIV patient who underwent a total cystoprostatectomy with ileal neobladder. The progression of glandular cystitis is controversial, and the question of the risk of malignant degeneration is under discussion. Recurrences are frequent, occurring continuously or intermittently with variable intervals. After transurethral resection of the prostate (TURP), annual surveillance with cystoscopy and bladder biopsies is recommended [18].

CONCLUSION

Intestinal glandular cystitis is a rare condition affecting primarily men and involving the bladder mucosa. Its symptoms are nonspecific and often go undiagnosed. Transurethral bladder resection remains the treatment of choice, but other therapeutic modalities, proven effective, should not be dismissed. The issue of malignant degeneration remains a controversial topic for debate, but the course of the disease is primarily marked by recurrences, necessitating cystoscopic monitoring.

Conflict of Interest

The authors declare no conflicts of interest.

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