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CASE REPORT

Xanthogranulomatous orchitis combined with contralateral acute pyogenic orchitis: A rare case report

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Abstract: Xanthogranulomatous orchitis is a rare, chronic, non-neoplastic inflammation. This report discusses a 79-year-old male patient presenting with bilateral scrotal swelling and dysuria lasting two weeks. Despite two weeks of antibiotic treatment, symptoms persisted, prompting transurethral prostate resection and bilateral orchiectomy. Postoperative pathology confirmed xanthogranulomatous orchitis on the left and acute pyogenic orchitis with hemorrhage on the right. Three months post-surgery, the patient reported satisfactory outcomes without complications.

Keywords: xanthogranulomatous orchitis, acute pyogenic orchitis, yellow granulomatous orchitis

1 Introduction

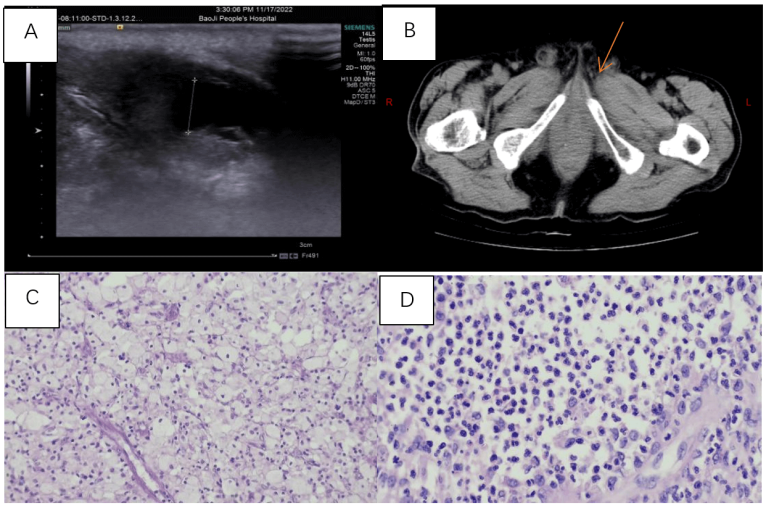
Xanthogranulomatous orchitis (XGO) is a rare, non-neoplastic condition characterized by nonspecific testicular inflammation, commonly presenting with pain and swelling. Surgical intervention is often necessary due to the ineffectiveness of medical management. This case illustrates a unique instance of xanthogranulomatous orchitis alongside contralateral acute pyogenic orchitis, previously unreported in literature.

2 Case report

Two weeks ago, a 79-year-old male manifested with bilateral scrotal edema and pain accompanied by dysuria. The patient had a medical history of chronic bronchitis and emphysema, and had undergone transurethral resection of the prostate two years ago. During the physical examination, bilateral scrotal enlargement with tenderness was observed, and a digital rectal examination indicated an enlarged prostate of degree III, with qualitative toughness and no significant induration. The laboratory examination results showed that TPSA was 7.30 ng/ml, FPSA was 0.639 ng/ml, carcinoembryonic antigen was 4.35 ng/ml, alpha-fetoprotein was 5.26 ng/ml, testosterone was 12.22 nmol/L, and lactate dehydrogenase was 106 U/L. The urine routine indicated that WBC was 2299/ul and RBC was 639.10/ul. No HCG was collected after admission. The scrotal B ultrasound revealed a mixed mass in the left testicle, which might be an inflammatory mass with a small amount of pus. The right testicle was swollen, but no abnormality in color flow was detected. The image in Figure 1A shows a fluid surround around the right testis. An MRI conducted at a different hospital revealed prostate enlargement that extended upward into the bladder, with a size of approximately 72.2 mm × 68.2 mm × 76.8 mm. A urinary CT scan indicated that the left testis had reduced in volume and had uneven density, while the right testis was enlarged. Inflammatory changes were also observed, as shown in Figure 1B.

After the patient underwent anti-infection treatment in our department, the anticipated effect was not attained. Despite the treatment, the patient's scrotal pain did not exhibit significant alleviation, and he persisted in experiencing dysuria. After thorough communication with the patient and his family members, we decided to carry out a transurethral prostate resection along with bilateral testicular exploration. During the operation, we discovered bilateral partial testicular tissue necrosis with a pungent odor and purulent discharge. After careful deliberation, we determined that conducting a bilateral orchiectomy was the most appropriate course of action. Postoperative examination disclosed that the left testis had xanthogranulomatous orchitis and local hydrocele alterations (Figure 1C), while the right testis had acute suppurative inflammation with hemorrhage (Figure 1D). Immunohistochemical results indicated that CD68 was positive,

while PSA and CK7 were negative, and Ki-67 had scattered positivity. Following a 3-month follow-up after the surgery, the patient had satisfactory postoperative outcomes and experienced no adverse events.



Note: A: scrotal B ultrasound: the mixed mass in the left testicle considers the inflammatory mass and a little pus. Bilateral seminal cord vein internal diameter thickening, echo enhancement, consideration: inflammatory changes; **B:** Urinary tract CT: left testicular atrophy, right testicular swelling, considering inflammatory changes; **C:** Pathological map of granulomatous orchitis (left): sheet distribution of foam tissue cells. (× 100); **D:** Pathological map of pyogenic orchitis (right): large amount of neutrophils, small amount of lymphocytes, plasma cells and histiocyte infiltration. (× 400).

Figure 1 The scrotal B ultrasound, MRI, and Pathological map

3 Discussion

Xanthogranulomatous inflammation (XGI), initially described by Wiener et al. in 1987, is a rare non-neoplastic disorder characterized by nonspecific inflammation. This condition predominantly affects men aged 50 to 70 years, with an incidence ranging from 0.6% to 1.4% [1–4]. It is most frequently identified in the kidneys and gallbladder, but it can also involve the liver, appendix, ovaries, vagina, bones, and bladder. When it invades the testis, it is known as testicular anthogranuloma (XGO). A case of yellowish granulomatous orchitis with contralateral acute suppurative orchitis is presented here. This is a rare benign testicular disease characterized by necrosis of testicular tissue followed by infiltration of lipid-filled macrophages [5–7]. The microscopic analysis of XGO indicated an accumulation of foamy tissue cells along with mixed heterogeneous inflammatory cells, resulting in significant damage and loss of the testicular parenchyma (Figure 1C). Widespread mixed inflammatory cell infiltration of the testicular parenchyma, testicular web, and epididymal tissue, with complete destruction of seminiferous tubules, fluid necrosis, suppuration, extensive fibrosis, and aggregation of foamy histiocytes with multinucleated giant cells [8, 9].

Immunodeficiency, persistent infection, and abnormal phagocytosis of necrotic tissue are all possible causes of xanthogranulomatous lesions [10, 11]. The pathophysiology of xanthogranulomatous orchitis is still unknown [12]. It is believed that XGO is linked to testicular chronic inflammatory infection and the ischemia process of the genital tract. Another cause of xanthogranulomatous testicular epididymitis is urine reflux via the vas deferens [10]. Salako et al. (2006) [13] consider Adult testicular xanthogranuloma is thought to be caused by epididymis or spermatic cord obstruction, which causes sperm extravasation and stasis. Given that the urinary B ultrasonography and MRI show aberrant prostate volume expansion (72.2 mm × 68.2 mm × 76.8 mm), we suspect that lower urinary tract obstruction is the root cause of this patient's xanthogranulomatous orchitis.

The most common differential diagnosis of xanthogranulomatous orchitis is acute orchitis and testicular tumor [14, 15]. Their common clinical manifestations were all those of scrotal enlargement. Common acute orchitis often has a history of acute infection, accompanied by scrotal redness and heat pain; testicular tumors generally have no obvious pain, often accompanied by obvious scrotal swelling, some patients can help serum tumor markers. Our case had atypical clinical features, only bilateral scrotal pain, negative testicular tumor markers, and an onset of 2 weeks, which could not be easily distinguished from common orchitis.

Currently, XGO has been reported to coexist with testicular tumors. Val-Bernal et al. [11] once reported a 52-year-old man who underwent orchiectomy for unilateral testicular enlargement and scrotal skin sinus formation, and the postoperative pathology suggested yellow granulomatous testicular epididymitis with seminoma. Given this possibility, histopathology became the most reliable evidence for the diagnosis of XGO and the exclusion of occult malignancy. Some studies have reported that B-ultrasound-guided fine needle biopsy (FNA) has diagnostic significance for yellow granulomatous pyelonephritis and cholecystitis, and FNA is expected to become a useful tool for preoperative diagnosis of XGO [16], which may prevent orchiectomy due to unexpected diagnosis.The general management of xanthogranulomatous orchitis includes anti-inflammatory, analgesic, detumescent, and symptomatic treatment, but the effect is not ideal. Orchiectomy for Matsumura et al. (2016) [17] was ultimately chosen in many cases. In adolescents with testicular swelling and pain, anti-infective treatment is generally preferred.

This case suggests that in patients with acute and chronic orchitis, if there is lower urinary tract obstruction, if antibiotic treatment is ineffective, the possibility of xanthogranulomatous orchitis should be considered. This case has certain guiding significance for the follow-up peers to formulate timely and decisive treatment plans.

Ethical statement

According to our ethics review board, an ethics approval is not necessary for a case report; so ethical approval is not required for this study in accordance with local guidelines. All procedures performed in this case report were in accordance with the ethical standards of the institutional and/or national research committee and with the 1,964 Helsinki Declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. This retrospective review of patient data did not require ethical approval in accordance with local guidelines.

Conflict of interest

The authors have no conflflicts of interest to declare.

Author contributions

Conception and design: Yu Dai, Yan-hu Meng; Administrative support: Yingyi Li; Provision of study materials or patients: Shuangning Liu; Collection and assembly of data: Yu Dai; Data analysis and interpretation: Yan-hu Meng; Manuscript writing: All authors; Final approval of manuscript: All authors.

Data availability statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

References

[1] Chung MK, Seol MY, Cho WY, et al.: Xanthogranulomatous cystitis associated with suture material. Journal of Urology. 1998, 159(3): 981-982. https://doi.org/10.1016/s0022-5347(01)63794-7

[2] Repetto P, Bianchini MA, Ceccarelli PL, et al. Bilateral xanthogranulomatous funiculitis and orchiepididymitis in a 13-year-old adolescent boy. Journal of Pediatric Surgery. 2012, 47(10): e33-e35. https://doi.org/10.1016/j.jpedsurg.2012.05.011

[3] Liang L, Jiajia W, Shoubin L, et al. Granulomatous orchitis: case report and review of the literature. Journal of International Medical Research. 2021, 49(5): 030006052110037. https://doi.org/10.1177/03000605211003773

[4] Wiener LB, Riehl PA, Baum N. Xanthogranulomatous Epididymitis: A Case Report. Journal of Urology. 1987, 138(3): 621-622. https://doi.org/10.1016/s0022-5347(17)43279-4

[5] Bapir R, Ali RM, Hussien DA, et al. Xanthogranulomatous epididymo-orchitis: A case report with review of the literature. International Journal of Surgery Case Reports. 2022, 92: 106832. https://doi.org/10.1016/j.ijscr.2022.106832

[6] Gongora e Silva RF, Pinto IC, Constantino ECN, et al. Xanthogranulomatous orchitis: case report of a rare condition. AME Case Reports. 2019, 3: 4-4. https://doi.org/10.21037/acr.2019.01.03

[7] Sharma P, Ramteke P, Satapathy A, et al. Xanthogranulomatous Orchitis Presenting as a Scrotal Mass in an Elderly Male: Malignancy or Mimicker? Clinical Medicine Insights: Case Reports. 2019, 12: 117954761989029. https://doi.org/10.1177/1179547619890295

[8] Val-Bernal JF, Argueta L, Fernández-Flórez A, et al. Bilateral xanthogranulomatous orchitis in a tetraplegic patient. Pathology - Research and Practice. 2012, 208(1): 62-64. https://doi.org/10.1016/j.prp.2011.11.003

[9] Ezer SS, Oguzkurt P, Temiz A, et al. Xanthogranulomatous Orchiepididymitis: A Very Uncommon Cause of Scrotal Mass in Childhood. Urology. 2013, 82(1): 228-230. https://doi.org/10.1016/j.urology.2013.01.039

[10] Vaidyanathan S, Mansour P, Parsons K, et al. Xanthogranulomatous funiculitis and epididymo-orchitis in a tetraplegic patient. Spinal Cord. 2000, 38(12): 769-772. https://doi.org/10.1038/sj.sc.3101045

[11] Val-Bernal JF, González-Márquez P, Ramos-Barseló E, et al. Concurrent xanthogranulomatous orchiepididymitis and seminoma in the same testis. Pathology International. 2009, 60(1): 55-58. https://doi.org/10.1111/j.1440-1827.2009.02475.x

[12] Nistal M, Gonzalez-Peramato P, Serrano A, et al. Xanthogranulomatous Funiculitis and Orchiepididymitis: Report of 2 Cases With Immunohistochemical Study and Literature Review. Archives of Pathology & Laboratory Medicine. 2004, 128(8): 911-914. https://doi.org/10.5858/2004-128-911-xfaoro

[13] Salako AA, Olasode BJ, Eziyi AK, et al. Xanthogranulomatous orchitis in an adult Nigerian. International Journal of Urology. 2006, 13(2): 186-188. https://doi.org/10.1111/j.1442-2042.2006.01258.x

[14] Yamashita S, Umemoto H, Kohjimoto Y, et al. Xanthogranulomatous orchitis after blunt testicular trauma mimicking a testicular tumor: A case report and comparison with published cases. Urology journal. 2017, 14(3): 3094-3096.

[15] Hama amin Said S, Abdalla RY, Aghaways I, et al. Xanthogranulomatous orchitis: Review of the published work, and report of one case. Urology Case Reports. 2019, 27: 100908. https://doi.org/10.1016/j.eucr.2019.100908

[16] Vijayvergiya G, Gulwani HV, Ul Hasan S. Xanthogranulomatous epididymo-orchitis. Indian Journal of Urology. 2023, 39(1): 58-66. https://doi.org/10.4103/iju.iju_270_22

[17] Matsumura M, Taketani T, Horie A, et al. Pediatric granulomatous orchitis: Case report and review of the literature. Pediatrics International. 2015, 58(2): 155-158. https://doi.org/10.1111/ped.12749