

Inflammation and infection

Xanthogranulomatous orchitis: Review of the published work, and report of one case

Shakhawan Hama amin Said, Rezhin Yaseen Abdalla*, Ismaeel Aghaways, Ari Mohammed Abdullah

Sulaimanyah Teaching Hospital, 0046, Iraq

ARTICLE INFO

Keywords:

Xanthogranulomatous orchitis (XGO)
Xanthogranulomatous epididymo-orchitis (XGEO)
Transurethral resection of prostate (TURP)
Transurethral resection of bladder (TURBT)

ABSTRACT

Xanthogranulomatous reaction is rare begin disease affects many different organs, in clinical practice it is well known to affect kidney and gallbladder, but also affect male genital organs including (Testis, epididymis, and spermatic cord), we report a case 70-year old, diabetic patient presented with right scrotal swelling, had history of prior TURP, tumor markers was within normal range, ultrasound shows multiple hypo echoic lesions with moderate hydrocele, exploration done: pus found within tunica vaginalis, and destructed testicular tissue with necrosis, culture revealed (E.Coli), histopathology showed xanthogranulomatous orchitis.

Introduction

Xanthogranulomatous inflammation is a rare condition, it is benign process characterized by destruction and necrosis of normal tissue, then replacement of normal tissue with lipid-laden macrophages, microscopically characterized by prominent foamy macrophage infiltration, with a minor infiltration of lymphocytes and plasma cells, in contrast to other granulomatous diseases (multinucleated giant cells, and caseous necrosis) are not seen,¹ In the genitourinary system it may affect the (kidney, and bladder), in genital organs it may affects (testis, epididymis, spermatic cord, and prostate), it also well known that xanthogranulomatous pyelonephritis is much more commoner presentation in Urological cases, and it also may affect other organs like (appendix, gall Bladder, and ovaries), regarding review of published literature this disease mostly affects 3rd-7th decade of life, we take that to consideration that XGO, and XGEO are rare.

Case report

A-70 year old male patient presented with right scrotal swelling for about 1 month duration associated with fever, and rigor, he had prior history of TURP 4 months ago which was complicated with post operative right epididymo-orchitis, and retrograde ejaculation, he was treated conservatively on antibiotic, after 2 months another attack of epididymo-orchitis occurs, associated with fever and rigor, he was treated on oral antibiotic for 7 days but not responding, he is a known

case of diabetes mellitus, on oral hypoglycemic, and lipid lowering drugs, he had history of surgical procedure for his left femoral fracture following road traffic accident decades ago, he is retired and live a sedentary life style, neither smoker nor alcohol drinker, he is married, and completed his family, on examination: revealed distended and redness of right hemiscrotum, on palpation: enlarged, tender right testis, no palpable mass was defined.

He has leucocytosis 14×10^3 /micro liter, hemoglobin = 12.7 g/dl, Serum creatinine = 1.0 mg/dl, his random blood sugar was 275 mg/dl, he was poorly controlled diabetes, and he was completely on insulin injection during hospital stay, HbA1c level was = 11% (97mmol/mol), Ultrasound was done show multiple hypo echoic lesions suggesting multicoated right testicular abscess with moderate hydrocele, tumor markers was within normal range, exploration done, right testicular tissue necrosis, and pus within tunica vaginalis was found, pus send for culture and sensitivity it was demonstrating (E.Coli), and histopathological examination it revealed xanthogranulomatous orchitis.

Discussion

Xanthogranulomatous reaction is rare begin disease affects many different organs, like:gall bladder, appendix, in urology xanthogranulomatous inflammation is more frequently seen in kidney, genital organ involvement includes (epididymis, testis, spermatic cord, and prostate), in reported cases testis then epididymis are most commonly affected and involved by this pathology, solo involvement of spermatic

* Corresponding author.

E-mail addresses: drshakhawan82@gmail.com (S. Hama amin Said), rezheenmedicine@yahoo.com (R.Y. Abdalla), ismaeelagha@hotmail.com (I. Aghaways), ariabdullah1978@gmail.com (A.M. Abdullah).

<https://doi.org/10.1016/j.eucr.2019.100908>

Received 13 January 2019; Received in revised form 2 May 2019; Accepted 6 May 2019

Available online 07 May 2019

2214-4420/ © 2019 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

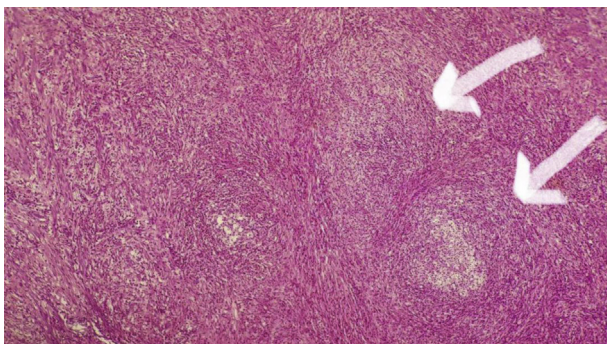


Fig. 1. Aggregates of foamy histiocytes forming non-caseating granulomas associated, with heavy mixed inflammatory cell infiltration, rich in lymphocytes and plasma cells, and fibrosis.

cord not reported yet, Process characterized by destruction of normal tissue which is replaced by lipid-laden macrophages, The etiology may includes (immunological defect, abnormal phagocytic activity on necrotic tissues, and chronic infectious conditions), despite correlated etiology pathogenesis of XGO, XGEO still not well established, chronic infection, and epididymal obstruction regarded to play a major role in pathogenesis of XGO, XGEO.

Spermatic tract obstruction is either Mechanical obstruction, or functional obstruction, spermatic tract dysfunction in case of diabetes caused by neuropathy also play a role which is reported in 6 cases,¹ Mechanical obstruction of the spermatic tract was reported in 4 case of post prostatectomy in patients with xanthogranulomatous orchioepididymitis,^{1,2} obstruction plays major role in pathogenesis but it is not a must, since there was a case reports that there is no clear cause for sperm flow obstruction like a case of XGO reported after intravesical BCG for case of bladder tumor following TURBT,³ or there was also a case report that both XGO and seminoma occurred in same testis.⁴

Infection also plays major role in pathogenesis, and initiating factor for xanthogranulomatous reaction, in all reported case where culture obtained majority demonstrate (E.Coli) infection, which clarifies that majorities of infections where local infection rather than haematogenous infection spread, which is due to obstruction or failure of ante grade sperm flow, and reflux of infected urine into reproductive system, and initiating immune response.

Differential clinical diagnoses of xanthogranulomatous orchioepididymitis include: bacterial infection (Usually caused by anaerobic infections and respond to antibiotic), testicular tumor (usually but not always associated with elevated tumor marker), Malakoplakia (presence of typical Michaelis–Gutmann bodies), since bacterial infection can respond to antibiotic and it can be differentiated from XGO during course of treatment and excluded from differential diagnosis, but in case of testicular tumor, and Malakoplakia exploration and excisional biopsy is mandatory for definitive diagnosis.⁵

Since XGO mimics testicular tumor, preoperative diagnostic tools like (testicular tumor markers, testicular and inguinal ultrasound, and fine needle aspiration) none of them give definitive diagnosis, Because of destructive nature of the disease, and massive tissue infiltration, for the definite therapy and diagnosis it brings surgery into mind by either complete or partial surgical excision.

Conclusion

Infection plays a major role in pathogenesis, obstruction and failure of antegrade sperm flow will initiate retrograde reflux of infected urine into reproductive system, infection will initiate immune response in reproductive system, immune response and macrophage infiltration plays major role in xanthogranulomatous reactions, since testis immunologically deprived by testicular blood barrier, so not all infection and obstructions will end up with immune response and

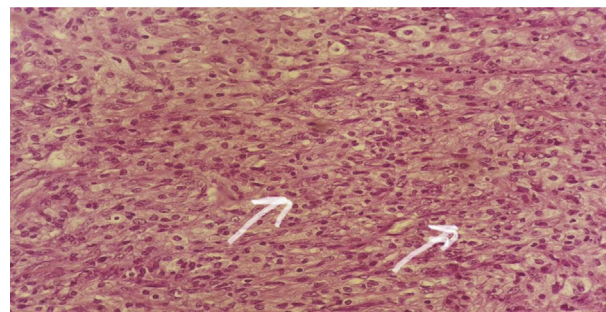


Fig. 2. Higher power view, showing foamy histiocytes.

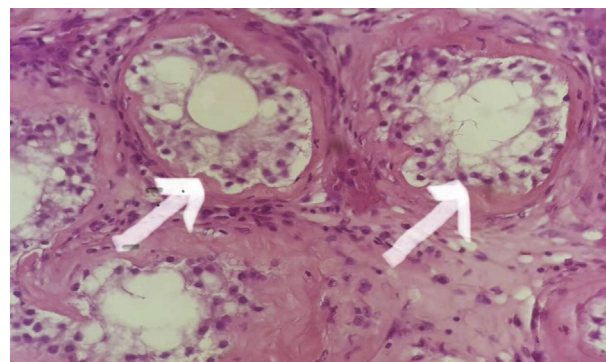


Fig. 3. Seminiferous tubules showing evidence hyalinization, lined by Sertoli cells with absence of germ cell.

xanthogranulomatous reaction.

Histopathology

In histological point of view granulomatous orchitis may be secondary to infection, trauma, autoimmune diseases, or extravasations of the sperm. Some of the specific causative agents include tuberculosis, syphilis, leprosy, and brucellosis. In many cases the cause remains unknown (idiopathic), in which distinct granulomas are not seen.

Xanthogranulomatous epididymo-orchitis is very rare, in our case the histological examination reveal well defined mass composed of aggregates of foamy histiocytes forming non-caseating granulomas (Fig. 1), higher power view shows heavy mixed inflammatory cell infiltration, rich in lymphocytes and plasma cells, and fibrosis (Fig. 2), the surrounding testicular parenchyma showing evidence of atrophy of the seminiferous tubule with marked thickening of basement membrane, the tubules were lined by Sertoli cells with complete absence of germ cells in most of them, and absence of active spermatogenesis (Fig. 3).

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.eucr.2019.100908>.

References

1. Nistal M, Gonzalez-Peramato P, Serrano A, et al. Xanthogranulomatous funiculitis and orchiepididymitis - report of 2 cases with immunohistochemical study and literature review. *Arch Pathol Lab Med.* 2004;128(8):911–914.
2. Alazab RS, Ghawanmeh HM, Al-Okour RK, et al. Xanthogranulomatous orchitis: rare case with brief literature review. *Urol Case Reports.* 2017;13:92–93. <https://doi.org/10.1016/j.eucr.2016.12.006>.
3. Hill JR, Gorgon G, Wahl SJ, et al. Xanthogranulomatous orchitis in a patient with a history of instrumentation and Bacillus calmette-guérin therapy. *Urology.* 2008;72(2):2007–2009. <https://doi.org/10.1016/j.urology.2007.12.007>.
4. Val-Bernal JF, Argueta L, Fernández-Flórez A, et al. Bilateral xanthogranulomatous orchitis in a tetraplegic patient. *Pathol Res Pract.* 2012;208(1):62–64. <https://doi.org/10.1016/j.prp.2011.11.003>.

5. Al-Said S, Ali A, Alobaigy AK, et al. Xanthogranulomatous orchitis: review of the published work and report of one case. *Int J Urol.* 2007;14(5):452–454. <https://doi.org/10.1111/j.1442-2042.2006.017>.