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Inflammation and Infection

Xanthogranulomatous Orchitis: Rare Case with Brief Literature Review



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ABSTRACT

Xanthogranulomatous reaction is uncommon non neoplastic reaction involving multiple organs, commonly kidney and gallbladder. Its rarely involve testicle. We report 69-year old man present to clinic with right testicular swelling for six month duration. Testicular markers within normal range. Us scrotum revealed right large avascular heterogeneous mass inside testicle. Right inguinal orchiectomy done and revealed Xanthogranulomatous orchitis.

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Introduction

Xanthogranulomatous inflammatory reaction (XGI) is an uncommon pathological diagnosis. XGI is a well-known disease entity of the kidney and gallbladder. However, XGI of the testicle has been rarely reported. To our knowledge, only few cases have been reported to date in the literature. Here, we present an additional case with brief literature review.

Case report

A 69-year-old man with history of TURP surgery 3 years ago, previously well, presented to our clinic with right scrotal swelling and a palpable mass of 6 month duration, which did not increase in size during this period. He was diagnosed outside our hospital with epidiymo-orchitis and received oral antibiotics for almost 3 months prior to presentation to our clinic without improvement He denied any urinary symptoms or previous similar problems in the past. On

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examination there was a right testicular firm mass with no tenderness over the testicle or the epididymis.

Testicular tumor markers and US scrotum were ordered. Tumor markers were within the normal range. US scrotum showed right heterogeneous hypo-echoic avascular mass, measuring about 2.5 cm \times 2 cm \times 2 cm, with normal epididymis (Fig. 1).

Patient was counseled about the possibility of testicular tumor or inflammatory reaction involving the right testicle and inguinal orchiectomy was advised. After informed consent, the patient underwent inguinal scrotal exploration with right orchiectomy. Histopathology assessment showed well circumscribed lesion composed of granulomatous inflammation. These granulomas were composed of foamy histocytes, macrophages and lymphocytes, with no evidence of malignancy. CD68+ was used to highlight the histocyte cell (Figs. 2, 3). The patient did not develop any post-operative complication during his hospital stay or follow up at the clinic.

Discussion

XGI is a rare inflammatory non-neoplastic disease that causes destruction and effacement of the normal structures of the affected organs. It has been a well-recognized disease entity in kidney and gallbladder.¹ However, it can occur in other organs like the liver, appendix, ovaries and urinary bladder. XGI also might affect the prostate, epididymis and testicle. XGI of kidney is found in only about 0.6–1.4% of all renal infections.^{2,3}

Statement of Originality: The material printed in this case report has not been previously submitted for another journal, and to the best of my knowledge contains no material previously published or written by another person except where due acknowledgement is made in the case report itself.

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Figure 1. Large heterogeneous avascular intra-testicular mass with areas of liquefactive necrosis.

The exact pathogenesis of xanthogranulomatous inflammation remains to be controversial. Advocate explanations suggest persistent chronic inflammatory process in the presence of partial or complete obstruction. This process is influenced by immunological defect of the macrophage, ⁴ persistent chronic infection, and foreign materials such as stone or retained suture material, ⁵ local response to tumor, abnormal lipid metabolism and lipid accumulation in the macrophages. ⁶

The suggested pathogenesis mostly comes from literature cases of XGI of the kidney as it is a well known disease and further investigations must be done to figure out the exact etiology of XGI of genital organs like testicle or epididymis.

On microscopic examination the yellowish nodules that contain lipid-laden macrophages mixed with inflammatory cells like lymphocytes, giant cells, and plasma cells might be surrounded by areas of necrosis. However, this not specific to XGI and might be present in various inflammatory or obstructive process.⁷

Differential diagnosis of XG orchitis includes testicular tumors, infectious epidiymo-orchitis, malakoplakia and idiopathic granulomatous orchitis.

In some cases, as the case presented above, it is hard to differentiate between XG orchitis and testicular tumors. Tumor markers

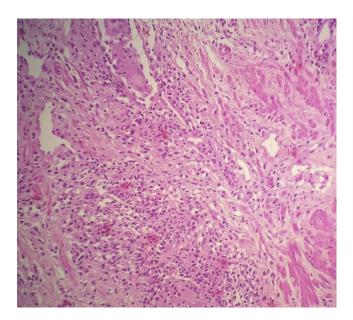


Figure 2. Severe inflammatory reaction with areas of necrosis adjacent to normal testicular tissue.

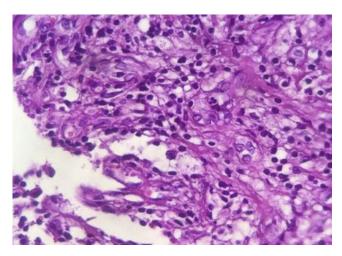


Figure 3. Higher magnification showing necrotic areas surrounded by inflammatory cells

might be helpful but many testicular tumors present with normal serum markers. Thus, careful history might be a good tool to guide physicians toward XG orchitis, especially in the presence of persistent infection, anatomical or functional abnormality of lower urinary tract. Nevertheless, inguinal surgical exploration still a wise decision in majority of cases.

Additionally, infectious epididymo-orchitis could improve by oral antibiotics. And malakoplakia can be distinguished histologically by "Michaelis—Gutmann bodies" which are associated with granular, eosinophilic macrophages.

Due to the destructive nature of XG orchitis, treatment should be aggressive by either radical or, in selected cases, partial orchiectomy.

Conclusion

XG orchitis is a rare disease. Few cases are reported in the literature. There is no definite radiological or laboratory method to diagnose this disease and it may present as testicular tumors. Curative treatment is by either radical or partial orchiectomy because of the aggressive nature of this disease. Further studies should be done to figure out the exact etiology of this disorder.

Conflict of interest

This case report was done in the division of urology for academic purposes and was not funded by any external fund, the submission has no commercial interests, and the authors of this case report are not linked to any external agencies.

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