Case Report

Xanthogranulomatous orchitis: case report of a rare condition

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Abstract: Xanthogranulomatous orchitis is a rare, non-neoplastic inflammatory condition. We present a case of a 55-year-old diabetic male with an increase in volume, pain and edema in right testicle with a one-year history. The examination involved the investigation of inflammatory markers and ultrasound. Exploration of the scrotum revealed important destruction of the tissue architecture. This case underscores the importance of including xanthogranulomatous orchitis in the differential diagnosis of a testicular lump.

Keywords: Orchitis; testicles, xanthogranulomatous inflammation

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Introduction

Xanthogranulomatous orchitis is a non-neoplastic inflammatory condition characterized by the destruction of testicular tissue with macrophage infiltrate (1). Xanthogranulomatous masses is often found in the kidneys and gallbladder, with few cases of occurrence in the testicle and spermatic cord (2,3). We report a case of xanthogranulomatous orchitis in an adult.

Case presentation

JMS, 55 years of age, diabetic and hypertensive, visited emergency care for testicular pain and swelling with an eight-day history, but a one-year evolution. The patient also reported having chills in the previous days and visited the emergency services on the seventh day of empirical treatment with ciprofloxacin for orchiepididymitis prescribed at another healthcare service.

The examination revealed testicular edema, hyperemia, increased volume with a hard consistency and pain to the touch of the right testicle as well as atrophy of the left testicle due to a past history of trauma.

The complete blood count revealed leukocytosis, increased segmented neutrophils and C-reactive protein of 13.28 mg/dL (value of reference: 0.00 to 0.50 mg/dL). Ultrasound of the scrotum revealed the right testicle with increase dimensions, absent vascular flow (Doppler ultrasound) and an adjacent pseudomass, with a thick epididymis, suggesting testicular torsion. The ultrasound exam revealed a hardened right testicle with no vascular flow, areas of necrosis and purulent secretion as well as thickening of the inguinal cord. Orchiectomy was performed with a margin up to the cord.

The patient received discharge from hospital on the second postoperative day. The biopsy revealed extensive testicular ischemic necrosis with areas of exudation, a chronic peri-testicular xanthogranulomatous inflammatory process, recent thrombosis, fibrous capsular thickening and acute epididymitis, with the formation of luminal abscesses confirmed by the immunohistochemical analysis (CD34-positive) (*Figures 1-3*).

The patient is currently in outpatient follow up, with occasional pain on the right side of the scrotum and undergoing hormonal replacement due to the atrophic post-trauma lest testicle.

Discussion

Xanthogranulomatous inflammation is rare with an unclear etiology. One explanation for this condition is a chronic

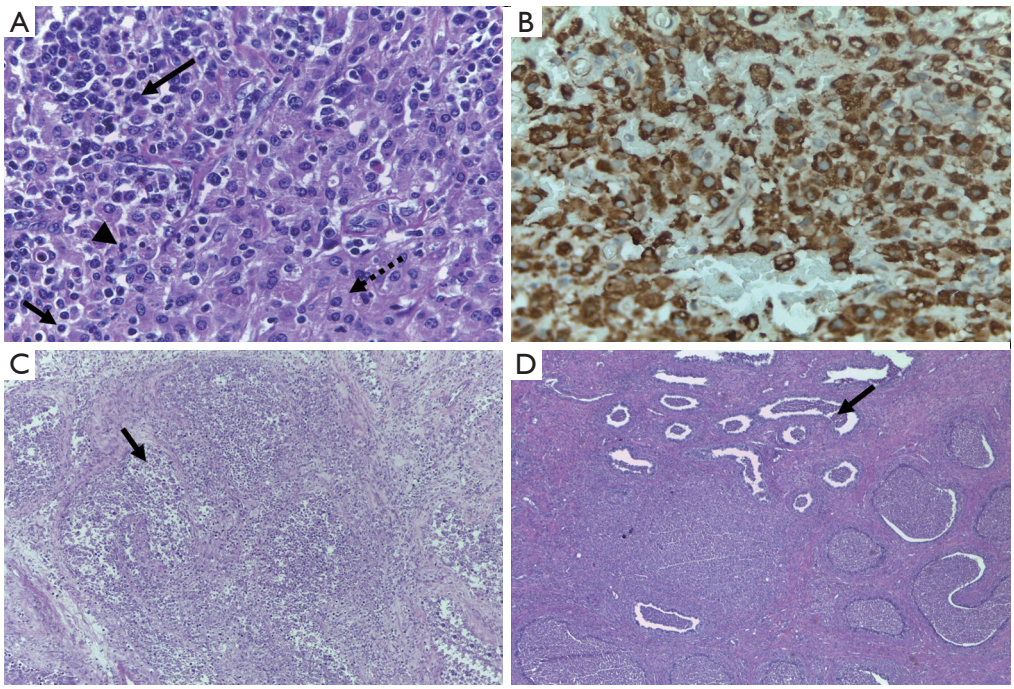

Figure 1 Hematoxylin-eosin staining and immunohistochemistry. (A) Hematoxylin-eosin staining showing chronic inflammatory infiltrate in testicular region composed predominantly of xanthomatous macrophages with central vesicular nucleus and eosinophilic cytoplasm with foamy appearance (dashed arrow) interlaced with neutrophils (head arrow), lymphocytes (short arrow) and plasmocytes (long arrow) (original magnification ×400). (B) Immunohistochemistry showing positivity for CD68 marker confirms histiocytic nature of xanthogranulomatous orchitis (original magnification ×400). (C) Hematoxylin-eosin staining showing Ischemic necrosis of seminiferous tubules associated with neutrophilic exudate (original magnification ×100). (D) Hematoxylin-eosin staining showing acute epididymitis with abscesses in epididymal lumen (original magnification ×100).

inflammatory process due to duct obstruction. This process is influenced by changes in the immune system and both lipid and macrophage metabolism, lymphatic dysfunction, a foreign body with calculus and surgical thread (4). Commonly found in the liver and gallbladder, xanthogranulomatous inflammation can also be seen in other organs, such as the appendix, ovaries and bladder (5), but can also affect the prostate, epididymis and testicle.

Diabetes, as in the present case, is directly linked to the etiology of xanthogranulomatous inflammation, since diabetic patients are often immunocompromised and the entire process of adherence, chemotaxis and phagocytosis is compromised (6).

Microscopic findings include yellowish nodules with "lipid-laden" macrophages associated to lymphocytes, giant cells and necrosis. However, such findings are not specific to the xanthogranulomatous process and could be concomitant to a tumor (7). The diagnosis is challenging, as the condition can resemble bacterial orchitis, which improves with antibiotic therapy, malakoplakia with typical Michaelis-Gutmann bodies (3) and neoplasm with differences in tumor and immunohistochemical markers (8).

Due to the destructive effects of xanthogranulomatous orchitis, treatment should be aggressive, with radical orchiectomy (as performed in the present case), which could be partial in some cases. Antibiotic therapy for anaerobic bacteria is also important (8-10).

As the patient became symptomatic due to the orchitis and atrophy, hormonal replacement therapy with testosterone was indicated to avoid hypogonadism (11).

Conclusions

Xanthogranulomatous orchitis is a rare condition with an unclear etiology. The present report serves as a reminder of the importance of this condition and can facilitate its understanding, differential diagnosis and the establishment of adequate treatment.

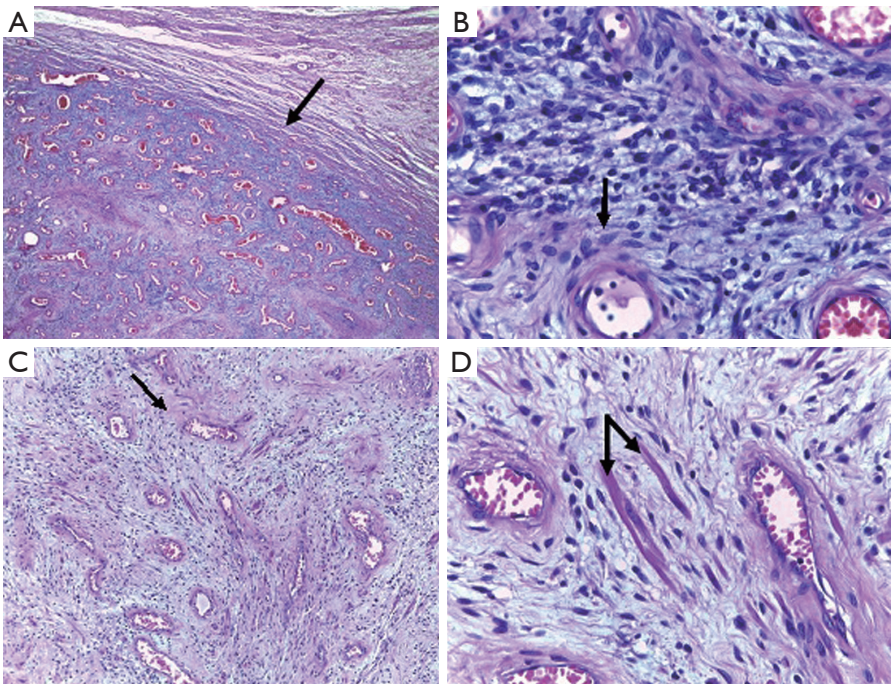

Figure 2 Hematoxylin-eosin staining showing: (A) well-defined mesenchymal growth located in testicular region lined by fibrous pseudocapsule (original magnification ×25); (B) tumor with proliferation of fusiform cells showing elongated nucleus with no atypias and scarce cytoplasm amid edematous stroma with prominent ectatic blood vessels of thin walls (original magnification ×400); (C) visible hypocellular areas contrasting with more cellular zones (original magnification ×100); (D) muscle cells found amid fusiform cells (original magnification ×400).

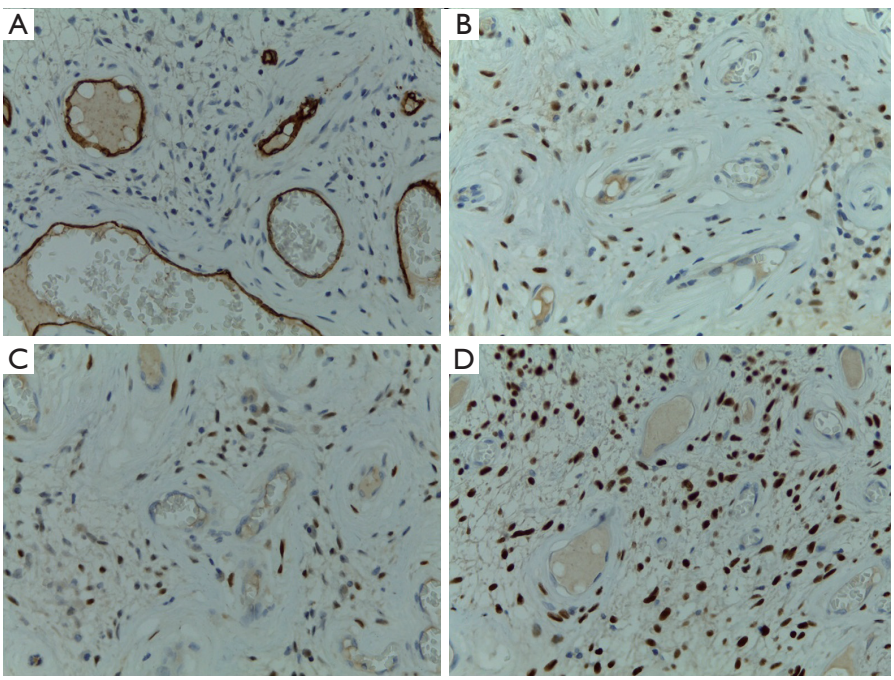

Figure 3 Immunohistochemistry showing negativity for CD34 marker in fusiform cells (A); nucleus positive for estrogen (B), progesterone (C) and androgen (D) receptors; positivity for CD34 marker in blood vessels (original magnification ×400).

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

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