

Inflammation and infection

## Xanthogranuloma of the epididymis

James Ting<sup>a</sup>, Qiuyu Jin<sup>a</sup>, Michael Biles<sup>b</sup>, Stephen A. Berry<sup>c</sup>, Maunank Shah<sup>c</sup>,  
Amin S. Herati<sup>b, 1, \*</sup>

<sup>a</sup> Johns Hopkins University School of Medicine, USA

<sup>b</sup> The James Buchanan Brady Urological Institute and Department of Urology, Johns Hopkins University School of Medicine, USA

<sup>c</sup> Division of Infectious Disease, Department of Medicine, Johns Hopkins University School of Medicine, Baltimore, MD, USA



### ARTICLE INFO

#### Article history:

Received 16 November 2017

Accepted 4 January 2018

Available online 9 January 2018

#### Keywords:

Adult

Humans

Epididymis

Tuberculosis

Epididymal surgery

Histology

### 2. Case presentation

A 66 year old man with HIV suppressed on antiretroviral therapy with a CD4 T-cell count of 470 cells/microliter, and a history of latent tuberculosis treated two years earlier with a 9 month course of isoniazid, presented to the urology clinic for evaluation of chronic bilateral scrotal swelling. The scrotal swelling progressively enlarged over a 10 year period. Scrotal duplex ultrasound (Fig. 1) revealed three solid, well-circumscribed right-sided epididymal masses, as well as bilateral hydroceles. These masses were heterogeneously hypoechoic with multiple tiny internal calcifications; the largest measured up to 2.5cm. No vascularity was observed within the masses. While the masses lacked vascular flow on Doppler ultrasound, epididymal malignancy could not be ruled out given the calcifications. Therefore, the decision was made to proceed to the operating room for definitive surgical management with either a partial epididymectomy or radical orchiectomy depending on intraoperative frozen section findings.

Once the right testicle and epididymis were delivered through an inguinal incision, a tourniquet was applied to the spermatic cord. A microsurgical technique was used to perform a partial epididymectomy with enucleation of all three epididymal masses. The largest mass was bivalved on the OR table, and tan-colored caseous material was produced (Fig. 2). The specimen was cultured for aerobic and anaerobic organisms and sent for frozen section analysis, which returned positive for inflammatory granulomas. At this point, it was decided that a radical epididymectomy was unnecessary and the tourniquet was removed. A bilateral hydrocelectomy was then performed. TB was not suspected at the time, and no tissue was sent for mycobacterial culture.

Pathologic analysis was notable for epididymitis with fibrosis and xanthogranulomatous inflammation (Fig. 3). Warthin-Starry stain, Von Kossa, Syphilis, and fungal stains were negative with no organisms identified. Additionally, no Michaelis-Gutmann bodies were visualized and acid fast smear did not detect acid fast bacteria.

### 3. Discussion

A xanthogranuloma is a benign, inflammatory process of non-

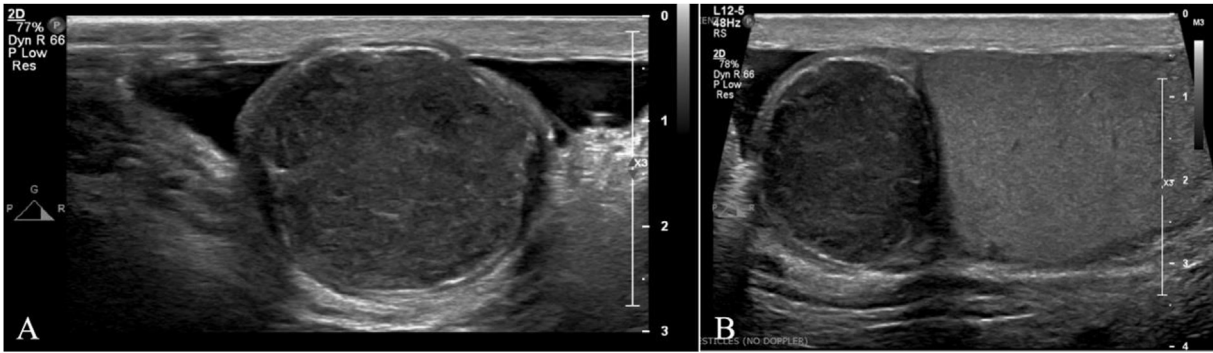
### 1. Introduction

Solid epididymal masses can be broadly classified into three categories: inflammatory mass, benign neoplasm, and malignant neoplasm. Examples of inflammatory processes include infectious epididymitis, fibrosis, sperm granuloma, and autoimmune disease. Benign neoplasms include adenomatoid tumor or leiomyoma. Malignant neoplasms include adenocarcinoma, embryonic rhabdomyosarcoma, and seminoma. While ultrasonography has shown promise in differentiating epididymal masses [1], a definitive diagnosis cannot be made without biopsy and histologic analysis. Reported worrisome signs suggestive of malignancy and indicating further exploration include large size and high vascularity on Doppler. [1] We describe a case of a 66 year old man with a history of HIV and latent tuberculosis (TB) who presented with chronic bilateral scrotal swelling.

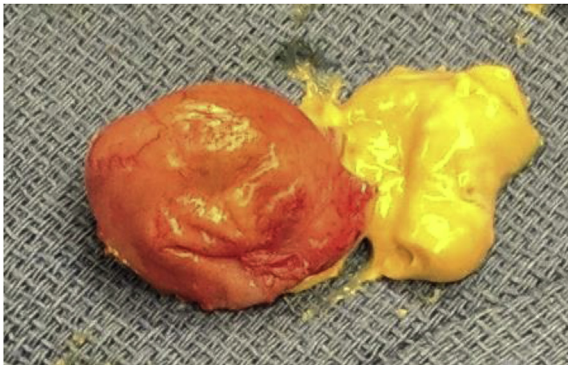
\* Corresponding author.

E-mail address: [aherati1@jhmi.edu](mailto:aherati1@jhmi.edu) (A.S. Herati).

<sup>1</sup> Assistant Professor of Urology, The James Buchanan Brady Urological Institute and Department of Urology, The Johns Hopkins University School of Medicine, 4940 Eastern Avenue, 301 Building, Suite 3112, Baltimore, MD 21224.



**Fig. 1.** a. Ultrasound image of circumscribed, peripherally calcified 2.5 cm × 2.4 cm × 2.3cm mass in the epididymal head. The mass is heterogeneously hypoechoic and contains multiple tiny internal calcifications. It does not demonstrate any internal color flow or invasive features. Two additional similar but smaller masses, not shown above, were also found in the epididymal tail. b. Ultrasound image obtained from the longitudinal axis of the right testis demonstrating a well circumscribed 2.5 cm mass in the head of the epididymis.



**Fig. 2.** Intraoperative photograph of epididymal mass measuring approximately 2.5cm and caseous exudate.

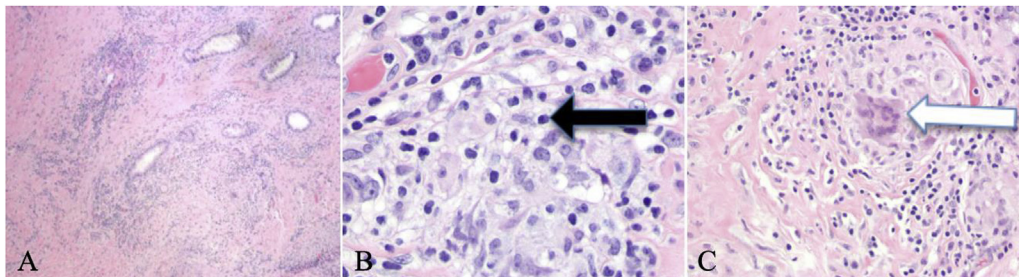
Langerhans cell histiocytes. In adults, it is most commonly found in chronic pyelonephritis. To our knowledge, only 20 or so cases have been reported that involve the male genital system, and here we present the first intraoperative image showcasing caseating exudate in an epididymal xanthogranuloma. [2,3]

Historically, xanthogranulomas of the genitourinary system were attributed to gram negative bacteria, when organisms could be isolated, [2] and usually in the context of local obstruction and

recurrent infections. This case was unusual as the patient had no prior history of bacterial urinary infections. While the acid-fast bacilli testing was negative, the sensitivity of acid-fast staining of formalin-fixed tissue compared to culture of fresh tissue is probably less than 50%. [4] With fresh tissue, an increasingly common adjunct to mycobacterial culture is TB nucleic acid testing. This is rapid compared to culture and may be 80% as sensitive. [5] The sensitivity of nucleic acid testing on formalin-fixed tissue is unknown but is likely lower, and we did not attempt it. We ultimately decided to treat this patient for presumed TB given his history of latent TB, the profound caseating material produced from the surgical specimen, and the patient's immunocompromised state.

#### 4. Conclusion

The workup of a patient who presents with an epididymal mass includes grayscale and color Doppler ultrasonography for initial characterization. If the masses cannot be sufficiently identified on imaging, a biopsy is necessary to rule out malignancy. In the case of a patient with HIV, it is also important for the clinician to maintain a high index of suspicion for tuberculosis as the etiology of an epididymal mass. In cases in which frozen section histopathology results are negative for malignancy, fresh tissue should be sent not only for bacterial staining and culture but also for mycobacterial staining and culture, and for TB nucleic acid testing if available. This case also demonstrates the unique histologic finding of



**Fig. 3.** Low-power and high-power pathologic sections showing an abundance of lymphocytes and multinucleated giant cells (C, white arrow). Foamy histiocytes (B, black arrow) were visualized, suggesting xanthogranulomatous inflammation.

xanthogranuloma in the context of probable tuberculous epididymitis.

#### Conflict of interest statement

Conflicts of interest: none.

#### Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

#### Appendix A. Supplementary data

Supplementary data related to this article can be found at

<https://doi.org/10.1016/j.eucr.2018.01.004>.

#### References

- [1]. Alleman W, Gorman B, King B, Larson D, Cheville J, Nehra A. Benign and malignant epididymal masses evaluated with scrotal sonography: clinical and pathologic review of 85 patients. *J Ultrasound Med*. 2008 Aug;27(8):1195–1202.
- [2]. Persec Z, Bulimbasic S, Persec J, et al. Xanthogranulomatous epididymitis: clinical report and immunohistochemical analysis. *Wien Klin Wochenschr*. 2008;120(11-12):355–369.
- [3]. Viswaroop B, Kekre N, Gopalakrishnan G. Isolated tuberculous epididymitis: a review of forty cases. *J Postgrad Med*. 2005;51(2):109–111.
- [4]. Fukunaga H, Murakami T, Gondo T, Sugi K, Ishihara T. Sensitivity of acid-fast staining for Mycobacterium tuberculosis in formalin-fixed tissue. *Am J Respir Crit Care Med*. 2002 Oct 1;166(7):994–997.
- [5]. World Health Organization. *Xpert MTB/RIF Assay for the Diagnosis of Pulmonary and Extrapulmonary TB in Adults and Children*. World Health Organization; 2013.