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Xanthogranulomatous Epididymo-Orchitis

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Xantogranulomatous epididymo-orchitis is an extremely rare inflammatory disease of the testis. The process can not be distinguished clinically from a malignant testicular tumor except on histological examination. To our knowledge there are only 18 cases reported in english literature.
A 34-year-old man presented with a month history of right testicular pain and swelling. Physical examination revealed severe tenderness with enlargement of the right hemiscrotum. Direct plain films and urinalysis were normal. Ultrasound examination of the right testis revealed non-homogeneous areas with poorly defined limits and hypoechoic-calcifying areas within the right testis and epididymis. A right testicular cancer was suspected and the patient had right inguinal orchiectomy. Microscopic examination showed diffuse replacement of seminiferous tubules and interstisium by sheets of foamy histiocytes intermingled with neutrophils, lymphocytes and plasma cells. Histochemical stains for organisms didn' t yield any fungi or mycobacteria. A diagnosis of xantogranulomatous epididymo-orchitis was made.
Key Words: Testis; Xantogranulomatous Inflammation; Differential Diagnosis.

Ksantogranulomatöz Epididimo-Orşit

Ksantogranülomatöz epididimo-orşit testisin, oldukça nadir görülen inflamatuar hastalığıdır. Bu durum, histopatolojik inceleme yapılmaksızın, klinik olarak malign testiküler tümörden ayırt edilemez. İngiliz literatüründe yayınlanan sadece 18 olgu mevcuttur.
34 yaşında erkek hasta, bir aydır devam eden sağ testiküler ağrı ve şişlik şikayetiyle başvurdu. Fizik muayenede sağ hemiskrotumda ciddi hassasiyet ve büyüme tesbit edildi. Radyolojik inceleme ve idrar analizleri normaldi. Ultrasonografide, sağ testis ve epididimde, sınırları belirsiz, homojen olmayan ve hipoekoik kalsifiye alanlar tesbit edildi. Sağ testis tümörü düşünülerek sağ inguinal orşiektomi uygulandı. Mikroskopik incelemede, seminifer tübüller ve interstisyumun, nötrofil, lenfosit ve plazma hücreleri ile karışık köpüksü histiositler ile yer değiştirdiği görüldü. Histokimyasal boyalarda mantar veya mikobakteri saptanmadı. Ksantogranulomatöz orşit tanısı verildi.
Anahtar Kelimeler: Testis; Ksantogranulomatöz İnflamasyon; Ayırıcı Tanı.

Introduction

Xantogranulomatous inflammation is an uncommon, non-neoplastic, distinctive type of chronic inflammation. It is characterized by destruction of the affected tissue by a cellulary infiltrate of foamy macrophages, dense lymphocytes and plasma cells. In the genitourinary system, xantogranulomatous inflammation is more frequently seen in kidney and bladder; however testicular involvement is a rare event. To our knowledge there are only 18 cases reported in the english literature.[1-8] We report an additional case and discuss the published data.

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Case Report

A 34 year-old man, presented with a 4 month history of right testicular pain and swelling. The patient reported

no prior history of urinary tract infection, orchitis, urolithiasis, trauma or diabetes. Physical examination revealed severe tenderness with enlargement of the right hemiscrotum. Direct plain films and urinalysis were normal. Urine cultures were negative. Laboratory results included a white blood cell count of $15.6x10^3$/M, a hemoglobin level of 15.8 g/dL.

Tumor markers αFP and βHCG were normal. Ultrasound examination of the right testis revealed non-homogeneous areas with poorly defined limits and also hypoechoic and calcifying areas within the right testis and epididymis. A right testicular cancer was suspected

and the patient had right inguinal orchiectomy. On gross examination of the specimen, the testis measured, 7.5x5x5 cm and its cut surfaces showed a white-yellow, non-circumscribed necrotic area that was 3.5 cm in diameter. Microscopic examination showed that the architecture of the testis was partially lost (Figure 1).

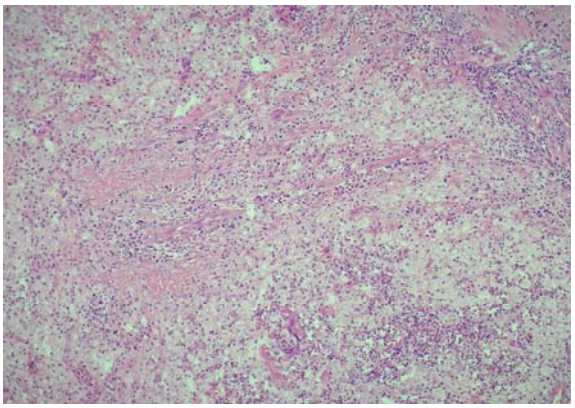

Figure 1. Xantogranulomatous inflammation infiltrating testicular parenchyma (H&E, x100)

There was diffuse replacement of seminifereous tubules and interstitium by sheets of foamy histiocytes intermingled with neutrophils, lymphocytes, mature plasma cells (Figure 2).

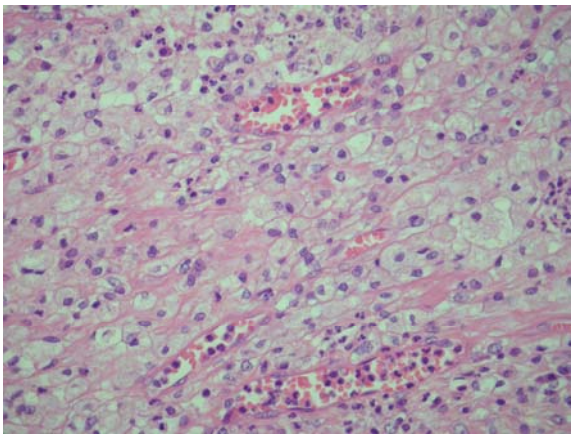

Figure 2. Xantogranulomatous inflammation is composed of foamy macrophages, dense neutrophils, lymphocytes and plasma cells (H&E, x200).

The inflammatory process also affected the epididymis. The histiocytic cells were positive for CD68 antibody. No Michealis-Guttman bodies were observed. Special stains for organisms (Acid-fast bacillus and Gomori Methenamine Silver) did not yield any mycobacteria or fungi. A diagnosis of xantogranulomatous epididymo-orchitis was made.

Discussion

Xantogranulomatous inflammation is an uncommon, non-neoplastic process characterized by the destruction of tissue which is replaced by cellular infiltrate composed of foamy macrophages, dense lymphocytes and plasma cells.[1-8] The most common genitourinary xantogranulomatous infection is pyelonephritis. It can also occur in other parts of the body such as the bladder, prostate and gallbladder.[2,3,5] However such inflammation in the testis, is not common with only 18 cases reported in English literature.[1-8]

Xantogranulomatous epididymo-orchitis can ocur in young adults and elderly. Presentation is unilateral in most cases. However bilateral cases have been reported. The right testis is commonly more affected than the left for unknown reasons.[7] The present case was a 34-year-old man and right testis was affected in accordance with the literature.

The etiology of xantogranulomatous epididymo-orchitis may include obstruction of the epididymis or ischemia of the testis and genitelia associated with chronic infection, urinary tract infections with ineffective antibiotic therapy, congenital urinary anomalies, altered immunological response, dyslipidemia and diabetes. Reflux of urine to the seminal vesicles or vas deferens and urethral manipulation (catheterization or surgery) were reported as possible risks for the development of the inflammation.[3-6] We could not find any possible etiological or risk factors in our case.

Xantogranulomatous epididymo-orchitis is difficult to diagnose preoperatively and can present in a variety of forms simulating other benign and malignant lesions of the testis. However the most feared is testicular cancer. Most cases mimic a testicular neoplasm clinico-radiologically. The testis is enlarged and firm to stony hard. Scrotal ultrasound is helpful in distinguishing other acute scrotal conditions such as torsion, but no specific ultrasound or magnetic resonance findings could distinguish xantogranulomatous epididymo-orchitis from testicular cancer.[3,5-8] Tumor markers can also be used for differential diagnosis but tumor markers may not be increased in some testicular tumors. In the present case, ultrasonography have failed to discriminate xantogranulomatous epididymo-orchitis from other pathological processes such as neoplasia and tumor markers were normal, so surgical exploration of the right testis was performed to establish the precise diagnosis.

Histologically, xantogranulomatous inflammation is characterized by destruction of normal tissue that becomes replaced by lipid laden macrophages and other inflammatory cells. Malakoplakia differs from

xantogranulomatous inflammation by the presence of typical Michaelis-Gutmann bodies associated with granular, eosinophilic and vacuolated macrophages. Differential diagnosis may also include specific orchitis. Epitheloid granulomas or caseous necrosis are absent in xantogranulomatous epididymo-orchitis.[5]

Because serious tissue destruction is a feature of xantogranulomatous inflammation, surgical removal is the curative treatment in most cases. Antimicrobial therapy for anaerobic infections are recommended as anaerobes may be important in the pathogenesis of xantogranulomatous inflammation.

In conclusion, xantogranulomatous epididymo-orchitis is a rare pseudoneoplastic condition and must be kept in mind in clinical and radiological differential diagnosis of testicular tumor.

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