Xanthogranulomatous Orchitis after Blunt Testicular Trauma Mimicking a Testicular Tumor: A case Report and Comparison with Published Cases

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Xanthogranulomatous orchitis has been reported to be an extremely rare inflammatory change caused by infection and is difficult to distinguish from testicular tumor. We report a 28-year-old man who presented with a lump in his left testis after a blunt testicular trauma. Based on a series of imaging tests, we suspected benign tumor such as epidermoid cyst and performed tumor enucleation (testis-sparing surgery) on the patient. Histopathological findings showed xanthogranulomatous orchitis. It is assumed that xanthogranulomatous orchitis in this case was caused by blunt testicular trauma and this is the first successful case of xanthoglomatous orchitis in salvaging the testis.

INTRODUCTION

Xanthogranulomatous inflammation is one form of chronic inflammation characterized by a cellular infiltrate of lipid-laden macrophages[1]. It is considered to be mainly caused by ascending or hematogenous infection, however clear pathogenesis is still unknown[2]. In the genitourinary system, xanthogranulomatous inflammation is commonly observed in kidney[3], bladder[4] and prostate[5]. It can also occur in the gall bladder[6].
Xanthogranulomatous orchitis (XGO) is a rare disease and, to our knowledge, only 22 cases have been reported to date. We present a case of XGO after blunt testicular trauma and compared with the published cases.

CASE REPORT

A 28-year-old man was hurt when his left testis was pinched in between the bicycle saddle and his left thigh. He had felt a dull pain in his left testis after the trauma, and a month later, he noted a lump in his left testis and visited our department. There was no history of urinary tract infection or urolithiasis. Physical examination revealed a thumb head size induration in his left testis. Complete blood count and testicular tumor markers were within normal ranges and urinalysis was normal. Various imaging tests, such as ultrasound, computed tomography and magnetic resonance imaging, showed a round tumor with distinct boundary between the tumor and the normal testicular tissue in his left testis (**Figure 1**). The right testis was normal.
Since we suspected benign tumor such as epidermoid cyst in this case, we tried to enucleate the tumor to preserve the testis. The tumor was easily separated from the normal testis. The surgery was completed once the tumor was

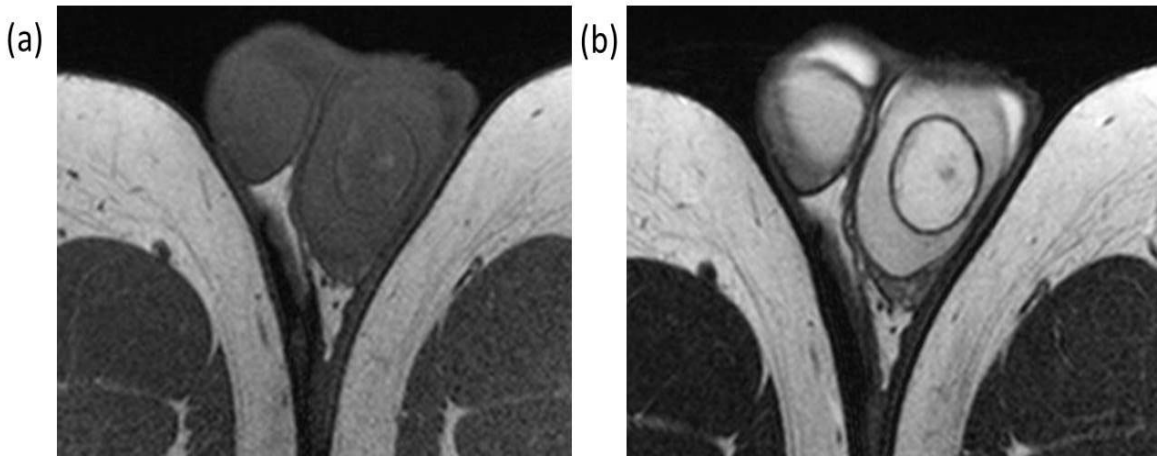

Figure 1. Magnetic resonance imaging showed a round tumor with distinct boundary between the tumor and the normal testicular tissue in the patient's left testis. **(a)** T1-weighted image. **(b)** T2-weighted image.

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Table 1. Summary of 22 published cases of xanthogranulomatous orchitis

Author	Journal	Total patients	Age	Diffuse or Focal	Infection type	Treatment
Akaza,H. et al.	JPN.J.Clin.Urol. 1997; 31: 1023-1026.	2	64,63	Diffuse	Ascending	inguinal radical orchiectomy in both cases
Iguchi,A. et al.	JPN.J.Clin.Urol. 1985; 31: 1023-1027.	1	51	Diffuse	Hematogenous	inguinal radical orchiectomy
Usamentiaga, E. et al.	Urology. 1998; 52: 891-892.	1	35	Focal	Ascending	inguinal radical orchiectomy
Vaidyanathan, S. et al.	Spinal Cord. 2000; 38: 769-772.	1	21	Diffuse	Ascending	inguinal radical orchiectomy
Hajiri, M. et al.	Ann.Urol(Paris). 2001; 35: 237-239.	7	30-75	Diffuse	Ascending	inguinal radical orchiectomy in all cases
Yap, R. L. et al.	Urology. 2004; 63: 176-177.	1	64	Diffuse	Ascending	inguinal radical orchiectomy
Nistal, M. et al.	Arch. Pathol. Lab. Med. 2004; 128: 911-914.	2	58,79	Diffuse	Ascending	inguinal radical orchiectomy in both cases
Dermirci, D.et al.	Int. J. Urol. 2004; 11: 686-688.	1	21	Difffuse	Hematogenous	inguinal radical orchiectomy
Salako, A. A. et al.	Int. J. Urol. 2006; 13: 186-188.	1	24	Diffuse	Ascending	inguinal radical orchiectomy
Al-Said, S. et al.	Int. J. Urol. 2007; 14: 452-454.	1	44	Diffuse	Hematogenous	inguinal radical orchiectomy
Hill, J. R. et al.	Urology. 2008; 72:461. e11-13.	1	68	Diffuse	Ascending	inguinal radical orchiectomy
Rifat Mannan, A. A. et al.	Med. Princ. Pract. 2009; 18: 418-421.	1	65	Diffuse	Hematogenous	inguinal radical orchiectomy
Val-Bernal, J. F. et al.	Pathol. Res. Pract. 2012; 208: 62-64.	1	55	Diffuse	Ascending	inguinal radical orchiectomy
Gakiya M. et al.	Int. J. Urol. 2013; 75: 90-94.	1	79	Diffuse	Hematogenous	inguinal radical orchiectomy
Present case		1	28	Focal		tumor enucleation

confirmed benign via rapid pathological diagnosis.
The resected specimen was a yellow necrotic mass with a diameter of 2 cm. Histological examination revealed the lesion consisted of a dominant infiltrate of foamy macrophages interspersed with a minor component of lymphocytes and plasma cells (**Figure 2**). Based on this inflammatory pattern, a diagnosis of XGO was made.

Discussion

We present a case of XGO which was caused by a blunt testicular trauma. **Table 1** summarizes the data of 22 published cases. Median age was 51.3 years old and XGO was considered to be caused by either ascending or hematogenous infection. The whole testis was replaced with fleshy tissue in 21 (95.5%) of the cases and inguinal radical orchiectomy was performed in all cases.

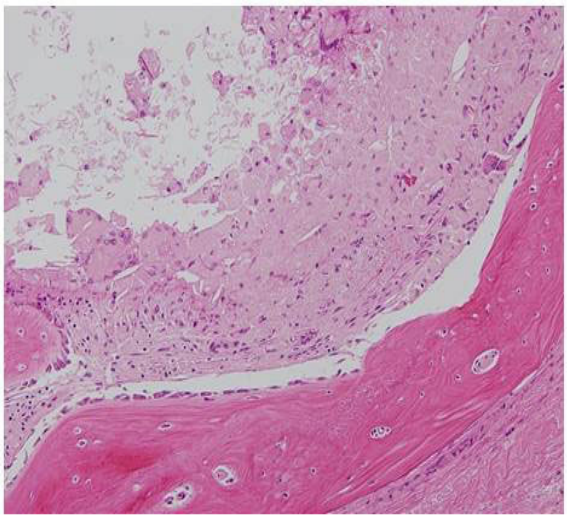

Figure 2. Foamy macrophages accompanied by lymphocytes and plasma cell infiltration. HE X 100.

In contrast, the present case differs from those previous cases. First, it is assumed that XGO in this case was caused by blunt testicular trauma as the patient experienced dull pain in his left testis since the trauma occurred and subsequently observed a lump a month after. All published XGO cases were caused by infection. However, in the present case, there was no sign of infection and urinalysis was normal. While there was a case of xanthogranulomatous pyelonephritis (XGN) which was reported to be caused by blunt trauma[7], the present case appears to be the first case of XGO caused by trauma. Although the exact mechanism of a trauma leading to xanthogranulomatous inflammation is still unknown, the following two hypotheses are considered. One hypothesis is that macrophages have infiltrated the hematoma caused by the trauma, resulting in XGO. The other hypothesis is that macrophages have infiltrated the necrotic tissue caused by blood vessel damage. The exact mechanism for causing xanthogranulomatous inflammation calls for future investigation.
Second, inflammatory tumor occurred in the normal testicular tissue and the patient underwent testis-sparing surgery. XGN accounts for lesser than 1% of chronic pyelonephritis and is classified into two categories. One is diffuse type in which the whole kidney is replaced with granulomatous tissue (about 85%). The other one is focal type in which granulomatous tissue forms a mass in normal renal tissue (about 15%)[3]. The present case suggested that there is also focal type of XGO. Almost all the cases of XGO (21/22 cases) were diffuse type where normal testicular tissues were not found.

Also, the patients underwent inguinal radical orchiectomy because it was difficult to rule out the possibility of malignant testicular tumor. However, although it was a focal-type XGO in the present case, the patient was initially diagnosed with epidermoid cyst, and was arranged to undergo tumor enucleation, also known as testis-sparing surgery. One focal-type XGO patient was reported to have underwent radical inguinal orchiectomy due to multiple tumors in his testis[8]. Therefore, to the best of our knowledge, this is the first successful case of XGO in salvaging the testis. The present case suggests that it is highly challenging to distinguish focal-type XGO from epidermoid cyst, which has relatively high generating frequency among benign tumors in the scrotum, based on radiological findings.

CONCLUSIONS

The present case suggested that trauma could cause xanthogranulomatous inflammation. As there is a focal type of XGO, it is therefore difficult to clearly differentiate it from epidermoid cyst. Hence, if epidermoid cyst is suspected, noting the possibility of a focal type of XGO and performing testis-sparing surgery should be taken into consideration.

CONFLICT OF INTEREST

None declared.

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