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A Giant Primary Sclerosing Lipogranuloma of the Scrotum

Skrotumun Dev Primer Sklerozan Lipogranülomu

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ABSTRACT

Sclerosing lipogranuloma is a rare, benign disease that can affect several organs, particularly of genitourinary system in males. The majority of the cases are secondary to exogenous foreign bodies. The masses are composed of granulomatous tissue formed around an either exogenous or endogenous lipomatous substance. We describe a 47-year-old male patient who presented with a growing, painless scrotal mass on physical examination. The mass was in 20 cm diameter and the laboratory findings were in normal limits. Pathologic evaluation confirmed the diagnosis of scrotal sclerosing lipogranuloma. To the best of our knowledge, this is the biggest scrotal sclerosing lipogranuloma case in the literature. We aimed with this presentation to keep in mind this benign lesion and also to assist the algorithmic approach.

Key Words: Granuloma, Scrotum, Sclerosis, Adipose tissue

ÖZ

Sklerozan lipogranüloma nadir ve birçok organı etkileyebilen benign bir lezyon olup, erkeklerde özellikle genitoüriner sistemi tutmaktadır. Olguların büyük çoğunluğu ekzojen olarak maruz kalınan yabancı cisimlere sekonder gelişir. Kitleler ekzojen ya da endojen lipomatöz bir yapının etrafında oluşan granülomatöz reaksiyon sonucu oluşur. Olgumuz 47 yaşında, ağrısız büyümekte olan skrotal kitleye sahip erkek hastadır. Fizik muayenesinde kitle 20 cm çapta ve laboratuvar bulguları normal sınırlarda olarak saptandı. Eksize edilen kitlenin patolojik değerlendirilmesi sonucu sklerozan lipogranüloma tanısı verildi. Bildiğimiz kadarıyla literatürdeki en büyük çaplı primer skrotal sklerozan lipogranülom olgusudur. Amacımız skrotal kitlelerde bu benign antitenin hatırlanmasını sağlayarak algoritmik yaklaşıma katkıda bulunmaktır.

Anahtar Sözcükler: Granülom, Skrotum, Skleroz, Yağ dokusu

INTRODUCTION

Sclerosing lipogranuloma (SLG) of the scrotum is relatively uncommon granulomatous condition presenting with subcutaneous mass. SLG was first reported by Smetana and Bernhard in 1950 (1). This benign lesion has previously been described as involving such structures as the scrotum, perineum, penis, and spermatic cord (2,3). SLG is described as primary when idiopathic in origin, and cases induced by injection of pathogenic materials such as paraffin and mineral oil are referred to as secondary (4). We report a case of giant scrotal SLG treated surgically, together with a review of the literature. Our case was a primary SLG with no history of exogenous foreign bodies.

CASE REPORT

The patient was a 47-year-old man. He had a growing, painless scrotal mass of 5 years duration. He did not have any trauma or an exogenous material injection to the scrotum. An irregular, painless, solid scrotal mass was revealed by the physical examination (Figure 1). There were no lymphadenopathy and also no lesion in bilateral testes, epididymis. Laboratory findings (whole blood count, blood chemistry, serum tumor markers) and roentgenologic examinations (chest radiography and computed tomography of the abdomen) were all within normal limits.

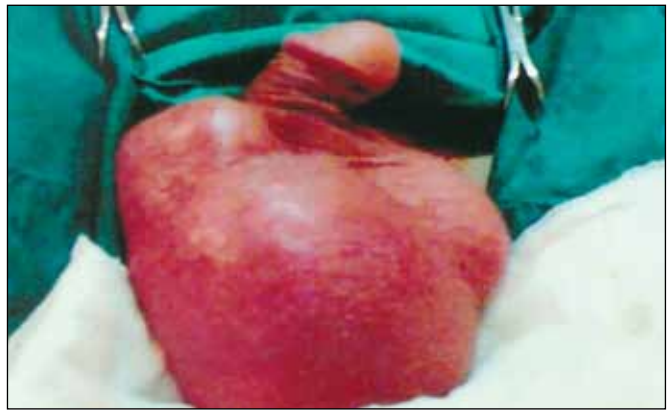

Figure 1: Macroscopic view of the scrotal mass.

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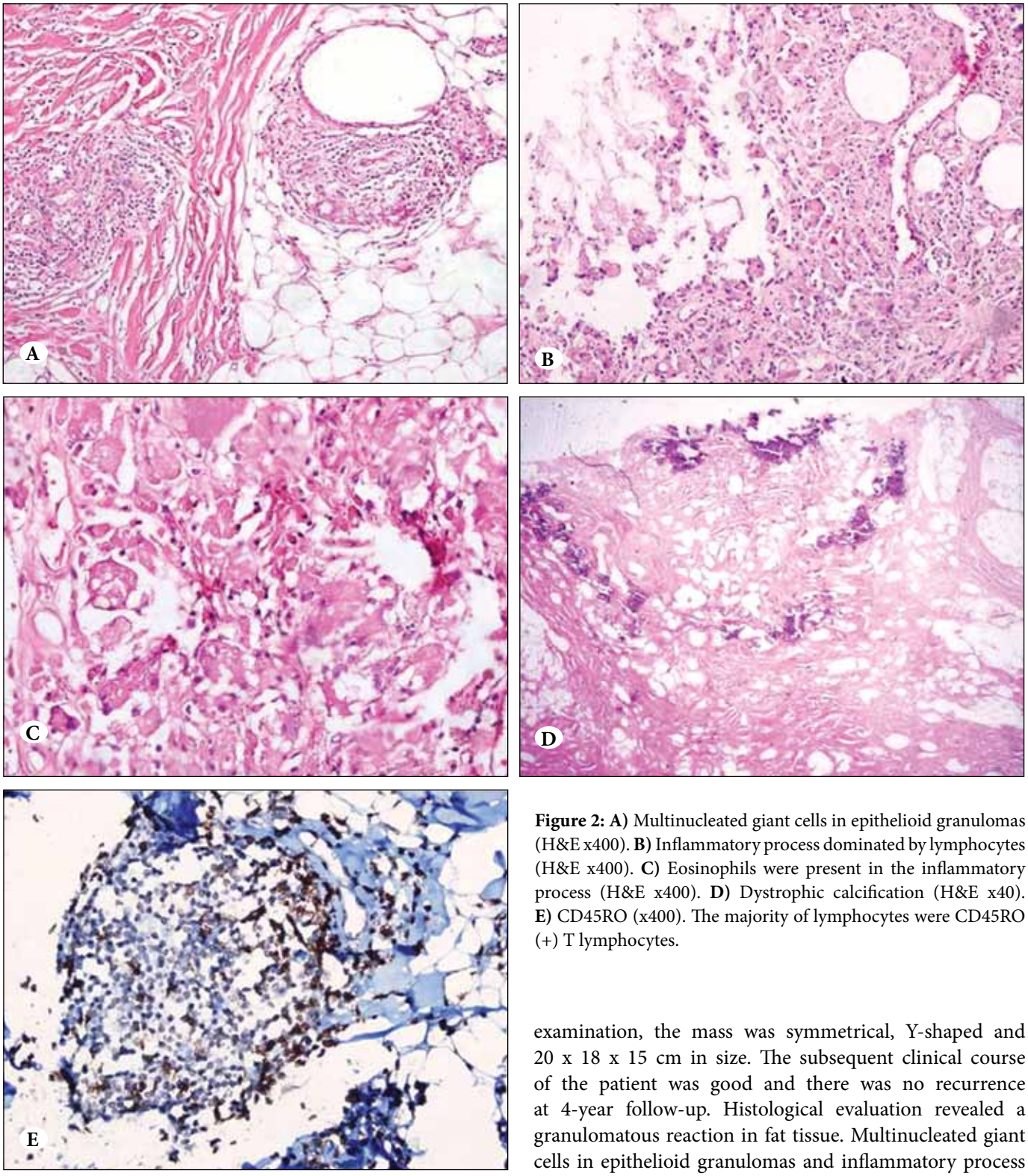

Figure 2: A) Multinucleated giant cells in epithelioid granulomas (H&E x400). **B)** Inflammatory process dominated by lymphocytes (H&E x400). **C)** Eosinophils were present in the inflammatory process (H&E x400). **D)** Dystrophic calcification (H&E x40). **E)** CD45RO (x400). The majority of lymphocytes were CD45RO (+) T lymphocytes.

The scrotal mass was resected, and the testicles, all adjacent structures were preserved. The mass was in the subcutaneous fat tissue and had no connection to the adjacent structures (testis, spermatic cord, epididymis, penis). At macroscopic examination, the mass was symmetrical, Y-shaped and 20 x 18 x 15 cm in size. The subsequent clinical course of the patient was good and there was no recurrence at 4-year follow-up. Histological evaluation revealed a granulomatous reaction in fat tissue. Multinucleated giant cells in epithelioid granulomas and inflammatory process dominated by lymphocytes were present (Figure 2A, B). Eosinophilic infiltrates were also present in the lesion (Figure 2C). In large sclerotic areas there were many foci of dystrophic calcification (Figure 2D), indicating that the mass was an old lesion.

An immunohistochemical examination was performed using Dako's ENVISION method. The primary antibodies used were CD45RO (Neomarkers, UCHL–1, ready to use), CD68 (Neomarkers, clone KP1, ready to use) and CD20 (Scytek, clone L26, ready to use). Most of lymphocytes (> % 95) were CD45-positive T cells, and 5% of lymphocytes were CD20-positive B (Figure 2E) lymphocytes. CD 68 was strongly positive in epithelioid cells and multinucleated giant cells. The result of polymerase chain reaction for mycobacterium tuberculosis was negative. Acid-fast bacteria were not identified by Ziehl-Neelsen stain. No fungi or gram-positive bacteria were observed using Grocott, gram, and PAS stains.

DISCUSSION

SLG was first reported by Smetana and Bernhard in 1950 (1). SLG of the male genitalia is a tissue reaction to exogenous foreign bodies such as paraffin, silicon (secondary SLG) or endogenous lipids (primary SLG) (3,4). Our case was a primary SLG with no history of exogenous foreign bodies. Histologically, SLG is a granulomatous lesion containing epithelioid cells, eosinophils, multinucleated giant cells, lymphocytes and macrophages (1, 2, 5).

The term eosinophilic SLG has also been used for this lesion because of marked eosinophilic infiltration and eosinophilia in peripheral blood. It has been suggested that it is closely related to the granulomas, although the mechanism remains unclear (6). Our case of primary SLG of the scrotum had mild eosinophilic infiltration (10%-15%) and no eosinophilia in peripheral blood. Immunohistochemically, the majority of lymphocytes were CD45RO-positive T cells, compatible with the hypothesis that degeneration of endogenous fat due to some allergic mechanism might be involved in the development of SLG (6). Microscopic examination revealed significant calcifications in the lesion. There has only been one previous study regarding bilateral SLG of the gluteal region with calcification, that by Iannello et al (7). In our case the dystrophic calcifications may be ascribed to the aging of the lesion over a 5-year period. At macroscopic examination the mass was symmetrical, Y-shaped and 20 x 18 x 15 cm in size. The SLG cases reported in the literature have measured between 1.5 and 9 cm. (7, 8). Our case is the largest scrotal SLG lesion in the literature. Tuberculosis, fungal infection and foreign body granuloma should be considered in cytological differential diagnosis of SLG.

A negative polymerase chain reaction for Mycobacterium tuberculosis was seen. No acid-fast bacteria were identified by Ziehl-Neelsen stain. No fungi or gram-positive bacteria were observed using PAS, Grocott and gram stains. The presence of the granulomatous reaction and the lymphocytic infiltration in particular led us to diagnose this benign lesion as primary SLG of the scrotum.

Definitive diagnosis can be established by histological examination of biopsied or resected specimens. Although steroid therapy has been recommended as the first treatment of choice, biopsy and surgical excision are frequently performed in the treatment of SLG (8, 9). Surgery should be the treatment of choice in patients with recurrence and in whom steroids are ineffective (8). SLG is a self-limited condition after biopsy in many reported cases (4, 6, 8,10). In the case of incomplete resection, however, the mass exhibits a rapid recurrence, thus mimicking a neoplastic lesion (6). No recurrence in our case was determined during the 4-year follow-up. In summary, we describe a very rare case of primary SLG of the scrotum. To the best of our knowledge, this is the largest example of scrotal SLG in the literature. Most of the reported cases in English literature patients are Japanese. We didn't have any knowledge about the demographical distribution.

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