Case Report

Surgical Management of Idiopathic Scrotal Elephantiasis: A Rare Case Report

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Abstract: Etiologically, massive scrotal lymphedema can be either congenital or acquired. Congenital is further divided into different types while the predominant variety in the acquired category includes infectious etiology. We present a case of a 35-year-old male presented with the complaint of scrotal swelling for 3 years. Blood tests for filarial, chlamydia, and tuberculosis were unremarkable. Excision of excessive scrotal skin was done and sent for biopsy. The wound was partially closed, and the remaining was left for healing by secondary intention. The biopsy report showed non-caseating granulomatous inflammation with no significant lymph node involvement.

Keywords: Scrotal elephantiasis, Lymphedema, Non-caseating granulomatous inflammation, Infectious etiology, Fibrosis, Hyperplasia.

INTRODUCTION

We present a case of a 35-year-old male presented with the complaint of scrotal swelling for 3 years. Scrotal lymphedema is caused by a blockage of the genital region's lymphatic flow resulting in swelling and edema. In addition to edema of the interstitial and subcutaneous tissue, there can be ultimate development of fibrosis, collagenous hyperplasia, loss of elasticity, and thickened skin. Etiologically, it can be characterized as congenital or acquired [1]. Sometimes no apparent cause is identified, and the syndrome is classified as idiopathic scrotal elephantiasis [2]. Of the acquired variety, it is usually seen due to infections caused by filarial infestation with *Wuchereria Bancrofti* [3], or *Chlamydia trachomatis* species [4]. Although usually not a life-threatening disease, it presents with significant physical and psychological morbidity and its complications include infection, scrotal abscess, or Fournier gangrene.

CASE REPORT

We present a case of a 35-year-old male with no known co-morbid who came via the outpatient department with the complaint of painless scrotal swelling for 3 years. Swelling was gradual in onset, initially small then gradually increased to the size of a bowling ball. For the first year, it was limited to the right side of the scrotum, after which it began to involve both sides and was associated with significant discomfort. Scrotal swelling was associated with penile swelling. There was no associated complaint of fever, burning micturition, dysuria, or hematuria. On genital examination, there was a scrotal swelling of approximately 35 x 25 cm in size which was soft in consistency, non-tender with overlying intact skin. Bilateral testes were not palpable. Transillumination test was negative. There was associated penile edema (Fig. **1A** and **1B**). The patient was admitted with a provisional diagnosis of scrotal elephantiasis and workup was done to determine the underlying etiology. Such etiologies include human immunodeficiency virus, tuberculosis, filariasis and testicular cancer, but all workup was unremarkable. The patient was planned for wide local excision for a better quality of life. The patient was placed in the supine position after spinal anesthesia and a wide local excision of the excess scrotal wall was done. Bilateral testes were separated from the underlying tissue (Fig. **2**). The scrotal wall excised, measured roughly 30 x 20 cm in size and weighed over 5 kg which was sent for histopathology. Due to the edema, plastic surgery advised leaving the surgical wound open for some time and then based on its healing, either attempting a delayed primary closure, or a possible full-thickness graft. Hence, with significant postoperative improvement at the surgical site, a partially delayed primary closure was done. A region of approximately 5 x 5 cm was left exposed for healing by secondary intention (Fig. **3**). The patient was discharged with the advice of daily dressing, good local hygiene, and fortnightly follow-up visits. The patient remained well postoperatively and was followed in our outpatient department after 2 weeks with the histopathology report which showed non-caseating granulomatous inflammation. Hence, the final diagnosis was idiopathic scrotal elephantiasis.

Fig. 1(A). Anterior View.

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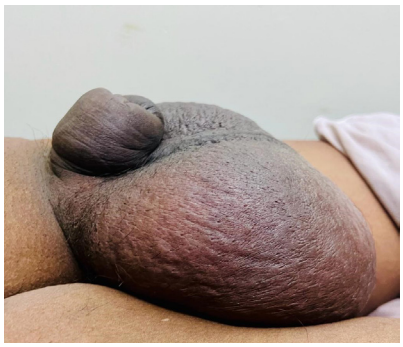

Fig. 1(B). Lateral View.

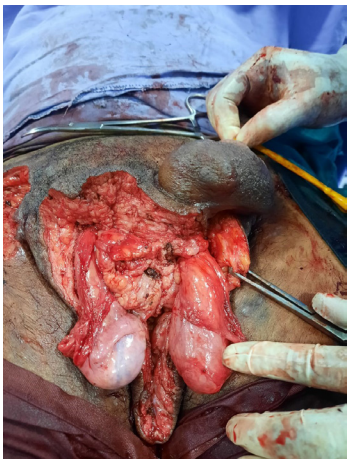

Fig. (2). Preoperative.

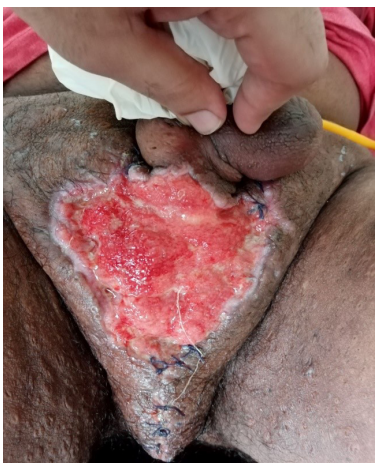

Fig. (3). Post-operative.

DISCUSSION

Scrotal Elephantiasis is a disease with documented cases dating as far back as 1859 [5]. However, till now it remains a rare disease. Over time, a better understanding of its etiology and corrective measures to tackle it has sprung up. Patients usually experience a sensation of discomfort and heaviness. The hyperkeratotic skin eventually develops into a sac containing exudative lymph in later phases. By allowing bacteria to enter the protein-rich lymphoid fluid in the sac due to the compromised epidermal barrier, cellulitis and complications such as abscesses, wound infections, and wound dehiscence may develop [6]. Therefore, if the patient presents late with significant signs and symptoms, surgical excision remains the main management. Surgical techniques usually involve reduction scrotoplasty which primarily rely on the presence of healthy scrotal skin that allow sufficient flap opposition and wound healing without involving complex rotational, advancement, or free flaps that rendered the healing questionable [7]. The patient's history favored an infective cause, however, the usual suspects' i.e. filarial infestations and chlamydia infections were ruled out by negative blood cultures. Other causes such as past surgical intervention, irradiation, or malignancy were also excluded. As per standard protocols and the patient's wishes, surgical intervention i.e., excision of the excessive scrotal wall was planned and executed. The primary aim was to improve the quality of life by limiting the scrotal and penile swelling and its resultant complications and decreasing the morbidity of the patient. However, due to the edematous nature of the skin, primary closure was delayed. The wound was closed partially and the remaining was left to heal using secondary intention. This plan of action went contrary to most cases in which the wound would be covered with a skin graft or rotational flap [2, 8]. Before choosing the best surgical strategy for the patient underlying cause, severity, and progress of the disease should be considered [9].

Based on the biopsy results, the patient was investigated for tuberculosis as a potential cause for his condition. Pakistan is known to have the fifth highest burden of the disease worldwide. According to a study, 22.7 % of extra-pulmonary tuberculosis cases had significant involvement in the lymphatic system [10]. In our case, since urine cultures, of acid-fast bacilli smear, and gene expert for acid-fast bacilli were negative, this could truly be designated as a case of idiopathic scrotal elephantiasis

CONCLUSION

Scrotal Elephantiasis is a rare disease that may manifest in all age groups though with varying etiologies. With better awareness, hopefully, such cases will be diagnosed earlier, hence leading to better outcomes with fewer surgical interventions needed for significant improvement in quality of life.

CONFLICT OF INTEREST

Declared none.

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Declared none.

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