

Genital Elephantiasis Surgery Treatment, A Case Report, and Literature Review

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Abstract

Introduction

Genital elephantiasis is a rare disease that causes pain, hygienic and social limitations. Surgical treatment aims to preserve function and improve genital esthetics.

Objective

To report a genital elephantiasis case submitted to genitoplasty and review the pathology and surgical treatment.

Method

The information was obtained through a review of the medical record, patient interview, photographic record of the pre, trans and postoperative period and research in periodicals.

Results

The case reported, and the literature has shown that the treatment of chronic genital elephantiasis associated with pain, mobility restriction or social disorder is essentially surgical. The procedure consists in excising all the tissue affected by the lymphedema sparing the posterolateral region of the scrotum for the scrotum reconstruction, followed by penile plastic with a flap of skin from the pubic region or graft. In the case reported, the patient was discharged on the second postoperative day and at five months of follow-up, there was a good aesthetic and functional result.

Conclusion

The surgical treatment of genital elephantiasis may be satisfactory in promoting patient rehabilitation.

Keywords: Elephantiasis; Genital Lymphedema; Surgical Treatment

Introduction

Scrotal elephantiasis (or massive scrotal lymphedema) is a disease caused by obstruction, hypoplasia or aplasia of lymphatic vessels that drain the scrotum [1]. It may be congenital or acquired: primary lymphedema is caused by an intrinsic defect of the lymphatic vessels [2]; secondary causes have as their origin surgical interventions, trauma, radiotherapy, malignant infiltration, venereal diseases and parasitic infections involving the inguinal lymph nodes, the best-known example is filarial infestation by *Wuchereria bancrofti* [3,4,5,]. Sexually Transmitted Infections (STIs) affects a significant range of patients that could develop genital elephantiasis, normally by Lymphogranuloma Venereum (LGV) and donovanose [6]. Giant scrotal lymphedema is relatively rare even in an endemic region for filariasis [2].

Since lymphatic drainage of the penis and scrotum predominates in the inguinal lymph nodes, elephantiasis only occurs when the chains are affected bilaterally or when a local factor, such as genital surgery, is involved [3].

This condition limits patients' mobility impairs urination and self-hygiene, and can determine fatigue, pain in soft tissue infected. It also causes restrictions on coitus, embarrassment and social isolation culminating with deterioration of quality of life [6].

Many cases can be treated conservatively, particularly if the primary disease is self-limited without permanent damage to the skin, lymph, and subcutaneous tissue. When lymphedema is chronic, permanent pathological changes that require resection of tissue to correct the disturbances are present [3]. Recently, a series of cases have been described that the treatment of surgical penoscrotal lymphedema through scrotal and penile plastic surgery provides the patient with more effective hygiene, with reduction of infections, ease of walking, orthostatic positioning, genital esthetic improvement and greater sexual satisfaction [1]. One of the most common techniques for treatment is resection of affected skin and subcutaneous tissue, preserving deep lymphatic drainage and reproductive structures, followed by reconstruction of the scrotum and penis [7].

Case Report

A 35-year-old male patient, from northeast of Brazil, with cognitive delay due to unclarified psychiatric affection, reported a progressive increase in scrotal volume started 8 years ago. Reports two unsuccessful surgical correction attempts. He reported scrotal pain, especially at the end of the day, difficulty in walking, urinating in the orthostatic position, performing penile hygiene practices, and difficulty in finding costumes that fit scrotal volume. Patient without a sexual partner. On examination, we found a testicular sac of 51 cm long and 64 cm in circumference, non-palpable testicles [Figures 1 and 2]. Ultrasonography study of the region revealed diffuse thickening of the scrotal wall suggesting lymphedema with topical testes in normal size and parenchyma. Based on the clinical and epidemiological history, all sexually transmitted diseases were tested and ruled out, in addition to the physical and imaging examination, the patient was initially diagnosed with genital elephantiasis; pharmacological treatment with diethylcarbamazine (6mg /kg/day) was instituted for 12 days and a decision was made for surgical treatment. The follow-up was 5 months until the writing date of this text.



Figure 1: Frontal view of genital lymphedema with patient in dorsal decubitus.



Figure 2: Lateral view of genital lymphedema with patient in dorsal decubitus.

For the surgical preparation, the abdominal region was cleaned to the knees with solution of chlorhexidine and trichotomy in the pubic, perineal and inguinal region [figure 3]. Bladder catheterization and prophylaxis with 2g of cefazolin was administered in anesthetic induction. The procedure began with the delimitation of the bilateral posterolateral wall grafts of the testicular sac and longitudinal incision in the median raphe of the scrotum from the base of the penis to the perineum and extending laterally to the base of the flaps [Figure 4]. The testicles with their respective spermatic strings were isolated and the vaginalis tunics were everted [Figure 5]. It was resected approximately 6 kilograms of the scrotal sac. Hemostasis of scrotal vascularization was performed, uniting medially the posterolateral flaps through suture to cover the spermatic cords and testes (aiming for the scar to simulate the scrotal raphe). The skin of the penis was excised above Buck's fascia to the balloonopreputial groove, ventrally, with the removal of excess skin and subcutaneous tissue and a pedicle graft of skin from the base of the penis to reconstruct the same, with a harmonious ending of the genitalia [Figure 6]. At the end of the surgery, the patient

was maintained with Foley's catheter and occlusive bandage in the penis and scrotum, that remained until the second postoperative day when the patient was discharged using scrotal suspensory.

The postoperative evaluation occurred weekly within the first month, then monthly until the 5th month, it was performed a subjective and comparative evaluation from the preoperative, observing a significant improvement of the aesthetic aspect of the external genitalia [Figures 7], improvement in behavioral and functional of the patient, such as urination, hygiene and ambulation. The anatomopathological result confirmed sclerosis and fusiform dilatation of lymphatic vessels. It was not possible to evaluate the sexual performance of the patient after treatment since he did not have a partner during the period followed. However, he reports masturbation more easily and with pleasure. Due to the daily hygiene and patient asepsis care, in the pre, trans and postoperative periods, no signs of infections in the region were identified, even though there is a risk for such.



Figure 3: Preparation of the patient for surgery.



Figure 4: Delimitation of the posterolateral grafts of the testicular sac.



Figure 5: Appearance after excision of the entire diseased testicular sac.



Figure 6: Final aspect of genital reconstruction.



Figure 7: Appearance after 5 months of surgery.

Discussion

In the case reported, we present an important lymphedema of the penis, scrotum and lower limbs. The history of chronic and progressive edema, as well as the study of epidemiological risks, patient's history, and complementary examinations, excluding others cited possible secondary causes, it could be inferred the diagnosis of secondary lymphedema caused by the parasite *Wuchereria bancroftii*, being pharmacologically and surgically treated for aesthetic and functional improvement of affected organs.

Genital lymphedema is an accumulation of lymph in the superficial lymphatic vessels between the skin and the fascia (Colles' fascia in the scrotum and Buck's fascia in the penis) [8]. Genital lymphoedema can be classified as primary and secondary [9].

Secondary lymphedema occurs when the lymphatic flow is interrupted or impaired due to lesion or resection of the lymph nodes, during surgical procedures, granulomatous disease, Paget's disease of the scrotum, Down's syndrome, infection, and radiation, among others [10]. In terms of lymphedema caused by parasites, the parasite *Wuchereria bancroftii*, which can affect up to 20% of the male population in tropical countries, is the most common [11].

In self-limited cases of filariasis and if there is no permanent damage to the skin, lymphatics and subcutaneous tissue, diethylcarbamazine is the treatment of choice for lymphatic filariasis. The treatment of monoinfection by lymphatic filariasis consists of diethylcarbamazine 6 mg/kg for 12 days. Alternatively, doxycycline (200 mg / d) may be added to the regimen for a course of 6 weeks [12].

The treatment of penoscrotal elephantiasis is essentially surgical. Several studies show the benefit of surgical treatment for the preservation of function and aesthetic improvement of the affected organ, including recent reports of techniques that showed little recurrence of the disease [1,8,13,14]. Surgery consists of a large excision of the subcutaneous tissue and skin affected by lymphedema, preserving deep lymphatic drainage, followed by reconstruction of the scrotum that is conventionally reconstructed using skin grafts [15], and penile restoration with a skin graft. The epididymis-testicular content is practically always respected [7].

The functional and aesthetic results are improved by the plastic reconstruction of a neoscrotum through posterolateral flaps made from the non-affected area and skin graft in the penile body giving it a normal appearance allowing the sexual act [13,14].

The excision of subcutaneous cellular tissue with preservation of the skin is a possibility, however, a review of the literature shows that only 1/3 of the patients are successful in the procedure while 2/3 require a new procedure or complete excision with cutaneous grafts [7,9]. Pedicled flaps of the thigh promote good aesthetic results, however, they are technically difficult and require that the disease does not affect the lower limbs [15], which would be impracticable for the case reported. The skin of the posterolateral region of the scrotum and perineum has been used for reconstruction since this region is not affected by the disease. This skin has collectorial lymphovascular drainage that ensures good results [1], with good appearance and it maintains the important thermoregulatory mechanism of the testes for spermatogenesis [16]. This technique was used in the case for the reconstruction of the testicular pouch.

Conclusion

Penoscrotal elephantiasis is a disease that compromises functionally and aesthetically affected patients. Although filariasis is relatively common in endemic regions, treatment is not always satisfactory. The plastic correction of this pathology enables the patient to return to a satisfactory condition.

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