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Idiopathic Penoscrotal Elephantiasis: A New Observation and Review of the Literature

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Abstract **Case Report**

Penoscrotal elephantiasis is a rare pathology outside filarial endemic areas. We report a case of primary penoscrotal elephantiasis in a 65-year-old patient, who was treated by complete surgical resection of the pathological tissue and penoscrotal reconstruction, with a good functional and aesthetic result. Complementary examinations and therapeutic options were analyzed through a review of the literature.

Keywords: Elephantiasis, Chronic lymphedema, Penis, Scrotum.

INTRODUCTION

Penoscrotal elephantiasis is a rare disease generally found in regions where filariasis is endemic [1-4]. It can reach monstrous sizes, which can be responsible for an unsightly appearance, a sexological impact due to burying of the penis, and psychological damage. It is diagnosed clinically and treated surgically. We report a new case of idiopathic penoscrotal elephantiasis treated surgically with a good evolution and a satisfactory functional and aesthetic result.

CASE REPORT

A 65-year-old man, circumcised at a young age and father of 4, consulted us with severe chronic lymphedema of the penis and bursa, which had been present for five years, interfered with walking and sexual activity and had no associated micturition disorders. We noted no history of a stay in a filarial endemic zone, scrotal trauma, pelvic surgery or radiotherapy or other history (heart failure, renal failure, hypoprotidemia). Clinical examination revealed penoscrotal elephantiasis, with bursae 65 cm in diameter and glove-finger-shaped burial of the penis with remodelled penile skin. The scrotal skin was thickened and cardboard-like (Figure 1). The rest of the examination was unremarkable. Blood tests for microfilariae and filariasis serology were negative. Chlamydia serology, performed as part of the search for lymphogranulomatosis venereum, was also negative. Abdominopelvic computed tomography revealed thickening of the scrotal walls, with no other detectable abnormalities. The diagnosis of primary scrotal elephantiasis was accepted and scrotal excision and plasty were performed. Surgery was performed in two stages: initially, the patient underwent resection of the elephantiasis scrotum, while healthy scrotal tissue was left at the bursal roots. This tissue enabled good-quality scrotal reconstruction, and the affected penile skin was also resected (Figure 2 and 3). The second stage of the operation was performed after 20 days and involved replacing the penile skin with a thin (7/10th mm) free skin graft taken from the thigh (Figure 4). Anatomopathological examination of the surgical specimen revealed extensive edema of the deep dermis and hypodermis, with dissociation of the scrotal muscle by edema and fibrosis, associated with an inflammatory infiltrate of the superficial dermis with orthokeratotic hyperkeratosis of the epidermal coating, suggestive of non-filarial scrotal lymphedema. The post-operative course was straightforward, and the patient was able to resume normal professional and sexual activity 4 months after surgery. The follow-up period was one year, with no local recurrence or notable incident.

Figure 1: penoscrotal elephantiasis: intraoperative appearance.

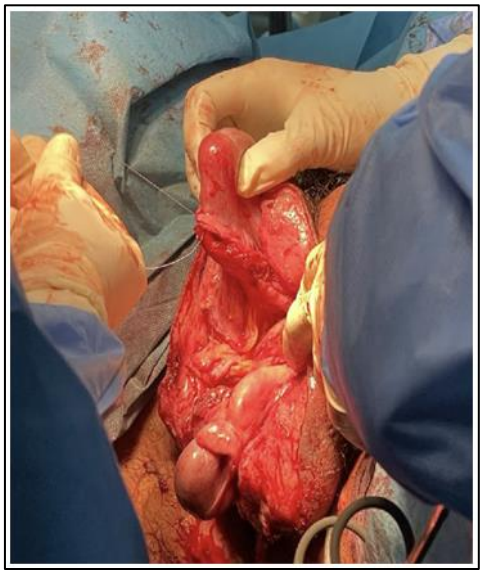

Figure 2: Complete resection of all elephantiasis tissue.

Figure 3: Result at the end of the first operation

Figure 4: Final result after penile skin replacement

DISCUSSION

Penoscrotal elephantiasis can be defined as an abnormal collection of protein-rich fluid in the subcutaneous tissue, due to a local alteration in oncotic or hydrostatic pressure. It is most often secondary to mechanical obstruction of lymphatic channels, either by inflammation and fibrosis, or by adult filarial worms [3, 4]. The most common cause of scrotal lymphedema is lymphatic filariasis. This parasitosis occurs mainly in tropical and sub-tropical countries [5], and mainly affects adult men in their forties [6]. Outside endemic filarial zones, scrotal elephantiasis may be congenital, secondary or, more often, idiopathic. Acquired causes are due to chronic mechanical or inflammatory obstruction, which may follow abdominal or pelvic tumors, carcinological surgery or pelvic radiotherapy [7, 8], Kaposi's disease, genital streptococcal or chlamydial infection [9], lymphogranulomatosis venereum [10], or hypoprotidemia [11]. Congenital or primary scrotal lymphedema is caused by a non-regressive dysplastic malformation of the lymphatic system, which is part of the lymphangioma family [11, 12]. Elephantiasis most often affects the scrotum or penoscrotal region, isolated penile involvement being rare, but the epididymo-testicular content is almost always respected [13]. In our case, the cause of elephantiasis remained undetermined. Scott McDougal has proposed a classification of these lymphedemas of the external genitalia according to congenital or acquired, sporadic or hereditary, and age of onset [14]. Isolated penile localization remains rare [5], and is confined to the penile skin and the glans penis [15], involvement is most often penoscrotal, generally sparing the epididymo- testicular contents [16]. In some cases, the penile skin is spared and involvement is purely scrotal [17]. Clinically, hypertrophy and deformity of the external genitalia are the main manifestations of elephantiasis of the external genitalia. Loss of elasticity of the skin, which thickens and takes on a cardboard-like appearance, has been consistently reported in the literature [18- 20]. Doppler ultrasonography is used to rule out an obstacle on the vascular axis of both lower

limbs. In cases of filariasis, it shows adult worms moving within the lymphatic vessels. Bipedal radiological or isotopic lymphography, which is used less and less frequently, sometimes shows pathognomonic aspects of lymphatic filariasis, with tiered lymphatic blockages, lymphangiectasias, granular and lacunar lymph node hypertrophies, sometimes with a "woollen skein" appearance. Lymphography can also be used to assess the possibility of surgical lympho-venous anastomoses. Magnetic resonance imaging provides a tissue characterization that correlates well with anatomopathological data, pinpointing the location of healthy tissue so as to limit surgical resection and facilitate scrotal reconstruction [21, 22]. With the exception of scrotal elephantiasis following radiotherapy for massive pelvic lymph node metastases, where conservative treatment is recommended due to the very limited life expectancy, treatment is based on surgery. It is undertaken after appropriate antibiotic therapy and stabilization of the inflammatory process [15- 21]. A wide exeresis of the pathological scrotal wall will be undertaken at the same time as a total superficial lymphangiectomy; it is recommended to start by freeing the spermatic cords and testes through an incision at the level of the superficial inguinal orifice [5]. Radical surgical excision is followed by scrotal plasty, which may involve several procedures: two bilateral scrotal flaps may be preserved, enabling reconstruction of a neo-scrotum. This technique is used by most authors and gives good results [6]. Inguinal or supra pubic pedicle skin flaps [5- 21]. The use of thin free skin has been described, but this could induce spermatogenesis disorders by altering testicular thermal regulation. For penile skin reconstruction, thin free skin grafts can be used, arranged in a spiral fashion to avoid retraction onto the penis [7- 23]. Sometimes, the extent of chronic lymphatic stasis can reach the spermatic cord and testicles, necessitating bilateral orchiectomy [10].

CONCLUSIONS

Penoscrotal elephantiasis is a rare condition, and the interest of this study lies in its therapeutic management. An etiological investigation is necessary before starting treatment. Surgical excision with scrotal plasty gives excellent functional and aesthetic results, enabling patients to return to a normal life.

CONFLICTS OF INTEREST : The authors do not declare any conflict of interest

CONTRIBUTION OF THE AUTHORS : The authors participated equally. All authors have read and approved the final version of the manuscript.

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