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Penoscrotal elephantiasis after treatment of prostate cancer: A case report

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Abstract

Penoscrotal elephantiasis (EPS) is a terminal state of lymphoedema defined by a progressive histopathological state characterized by chronic inflammatory fibromatosis of the subdermal and dermal connective tissue caused by lymphatic and venous stasis. Its clinical appearance is typical of an increase, sometimes considerable, in the volume of the external genital organs, which can lead to an unaesthetic appearance, a sexual impact, and psychological distress. The etiology can be primary or secondary to a parasitic disease (filariasis) or intrinsic or extrinsic lymphatic obstruction. Genital lymphedema may affect after cancer treatment and prostate cancer ranks as the second most prevalent cancer in men, making it crucial to consider any complications that may occur following its treatment.

We present a case of one patient, a 74-year-old man from Morocco, who underwent surgery two years ago for radical prostatectomy supplemented by radiotherapy and hormone therapy. The evolution was marked by an increase in scrotal size, with sexual and psychic prejudice. The Clinical examination revealed penoscrotal elephantiasis respecting the glans penis and both lower limbs. The diagnostic is clinical and etiological research requires specific complementary examinations, depending on the circumstances. Probably for this case, the penoscrotal elephantiasis was secondary to radical prostatectomy and/or post-radiotherapy. The patient was scheduled for surgical treatment involving en bloc resection of the elephantiasis scrotum, and immediate reconstruction after completely freeing the two testicles. The affected penile skin was also resected and replaced with a skin graft. The aesthetic and functional outcome was good with a follow-up of 7 months.

Keywords: Elephantiasis; Penoscrotal; Prostatectomy; Excision; Graft; Local flap.

1. Introduction

Elephantiasis is a terminal state of lymphoedema defined by a progressive histopathological state characterized by chronic inflammatory fibromatosis or hypertrophy of the subdermal and dermal connective tissue caused by lymphatic and venous stasis.

It distinguishes between primary or idiopathic lymphedema due to lymphangiectasia and secondary lymphedema due to chronic mechanical or inflammatory obstruction of the lymphatic system, which can have infectious, traumatic, post-radiation, or tumoral origins.

Peno-scrotal localization is less common than in the lower limbs and is clinically manifested by a large scrotum with a thickened or buried penis. This condition is noteworthy due to its significant functional, aesthetic, and psychological impact.

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We report the etiological, clinical aspects and therapeutic results of a case of penoscrotal elephantiasis following radical prostatectomy managed within our burns and plastic surgery department at the University Hospital of Tangier, Morocco.

2. Patients and observations:

This is a 74-year-old man from north of Morocco, a day laborer, who underwent surgery 2 years ago for radical resection of prostatic adenocarcinoma invading the left seminal gland, Gleason score 7(3+4), International Society Of Urological Pathology (ISUP) grade 2 with a prostate-specific antigen (PSA) of 21ng/ml supplemented by additional radiotherapy and put on hormone therapy based on Triptorelin 11.25mg injectable every 3 months. The evolution was marked by an increase in scrotal size, which interfered with walking and sexual activity, with psychic prejudice. In addition, there was no history of venereal disease, stay in a filarial endemic zone, or other antecedents (heart failure, kidney failure, hypoprotidemia).

Clinical examination revealed penoscrotal elephantiasis respecting the glans penis and both lower limbs (Figure 1). The rest of the examination was unremarkable.

Reno-bladder-prostate ultrasound with Doppler showed scrotal lymphedema corresponding to cutaneous and subcutaneous thickening of the scrotal lining reaching 20mm in thickness with no significant abnormalities in the testes or epididymides, and ruled out the presence of images of local recurrence, hydrocele or varicocele and ruled out testicular torsion.

A scrotal Magnetic resonance imaging (MRI) was performed to rule out local or locoregional recurrence and confirmed the presence of scrotal lymphedema, more marked on the left and extending to the perineal region, with no mass syndrome or associated testicular anomaly.

A prostate-specific antigen (PSA) test was requested as part of his follow-up, which was negative.

The diagnosis made was that of penoscrotal elephantiasis secondary probably to radical prostatectomy and/or post-radiotherapy.

The operative assessment was satisfactory, with hemoglobin at 14 g/dL, a normal coagulation profile, adequate renal function, and a normal ECG.

The patient was scheduled for spinal anesthesia for surgical treatment involving resection of the elephantiasis scrotum while preserving healthy scrotal tissue at the root of the scrotum (Figure 2). This tissue allowed for a high-quality and immediate scrotal reconstruction after completely freeing the two testicles. The affected penile skin was also resected and replaced with a split-thickness skin graft (7/10 mm) taken from the thigh. A silicone urinary catheter No. 18 was placed at the beginning of the procedure (Figure 3).

The surgical specimen weighed 740 grams and was sent to the pathologist. The histopathological examination of the specimen revealed significant edema of the deep dermis and hypodermis with dissociation of the scrotal muscle due to edema and fibrosis, associated with acute and chronic inflammatory changes in the superficial dermis and orthokeratotic hyperkeratosis of the epidermal surface, suggestive of non-filarial scrotal lymphedema.

The postoperative course was uncomplicated, and the patient was able to resume normal daily and sexual activities four months after the procedure (Figure 4). The follow-up was seven months without any local recurrence or notable incidents.



Figure 1 Penoscrotal elephantiasis: preoperative aspect.



Figure 2 Intra-operative view showing the liberation of the penis and both testicles.



Figure 3 Immediate postoperative aspect.

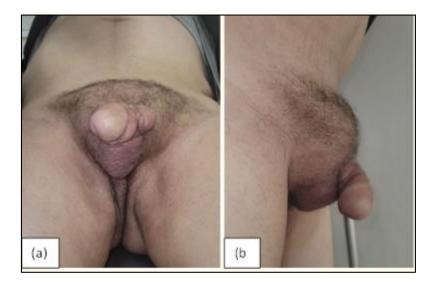


Figure 4 Final aspect at seven months follow-up with good healing and restoration of function and patient comfort; (a) inferior view, (b) profile view.

3. Discussion

We present the case of penoscrotal elephantiasis that occurred after a radical prostatectomy combined with adjuvant radiotherapy for prostate cancer. Given the rarity of epidemiological studies on this complication, estimating the incidence of penoscrotal elephantiasis post-prostatectomy remains difficult. Regardless of the incidence rate, penoscrotal elephantiasis deserves to be studied, as it can occur following the treatment of the second most common cancer in men and has a significant negative impact on the patient's quality of life.

Scrotal or penoscrotal elephantiasis is an increase in the volume, sometimes massive, of the scrotum with a typical clinical appearance. It is a rare condition outside countries where filariasis is endemic [1]. Outside these countries, it is more rarely primitive or idiopathic, caused by non-regressive dysplastic lymphangiectasia [2]. The scrotal lymphedema secondary to another etiology responsible for an obstruction of the lymphatic pathways which is due to an acquired condition of chronic mechanical or inflammatory origin such as surgical scar, abdominal or pelvic tumor, pelvic carcinological surgery, after radiotherapy, after infection (filariasis, streptococcal), sequelae of chronic venous stasis or surgery of urogenital bilharziosis, Kaposi's disease. [3]

Elephantiasis most commonly impacts the scrotum or the penoscrotal region, while isolated penile involvement is rare; however, the epididymis and testicular contents are nearly always preserved [4].

Epidemiological studies on genital lymphedema are limited, and most articles addressing secondary lymphedema after cancer treatment focus on its occurrence in the lower limbs, with very little detail on genital involvement. [5]

Secondary genital lymphedema can occur after various treatments for a wide range of cancers in the pelvic region and lower limbs (e.g., cancers of the uterus, cervix, ovaries, prostate, rectum, melanomas, Hodgkin or non-Hodgkin lymphomas), including surgical interventions with more or less extensive lymph node excision (inguinal, iliac, lumboaortic, pelvic), brachytherapy, and/or external radiotherapy [6,7].

Lower limb lymphedema after cancer treatment is poorly understood due to the lack of a consensual definition, with a wide range of frequency for a given cancer and its treatment, which often varies according to cancer classification, and it is rarely mentioned [8,9]

According to a retrospective study, 15 to 48% of women treated for gynecological cancer developed lower limb lymphedema, compared to 30% after melanoma or prostate cancer [7,8].

In the absence of specific questioning, and the difficulty of exploring the intimacy, the details regarding genital involvement are rarely mentioned. It is important to be aware that genital lymphedema is poorly understood by healthcare professionals.

In patients with lower limb lymphedema, the frequency of genital involvement is either impossible to determine due to the lack of specific questions or it appears more or less long after the onset of limb lymphedema, and the follow-up has been too short. The median time to consult a specialist after the onset of genital lymphedema after cancer treatment is 1.4 years [10].

Guidelines for prostate cancer recommend an extended pelvic lymph node dissection (ePLND) in conjunction with radical prostatectomy (RP) if the estimated risk of lymph node metastases exceeds 5% [11]. While women operated on for cervical cancer with ePLND report an occurrence of lymphedema of approximately 35% [12], versus a variant frequency of lymphedema after RP from 0% to 10% [13, 14], highlighting the need for a better understanding of the trade-off between the benefits and costs of lymph node dissection (LND) in prostate cancer.

The prospective controlled trial LAPPRO is a non-randomized multicenter trial including patients treated at 14 urological centers in Sweden. The trial includes patients 3675 men aged under 75 years who were operated on for localized prostate cancer. [15] Of the 3,675 men in the study, lymph node dissection (LND) was performed in 645 (17.6%). At 3 months after radical prostatectomy (RP), the prevalence of moderate to severe swelling in the groin and legs reported by patients was 13.7% among men who underwent LND, compared to 3.0% among men who did not. [16]

In patients with (LND), the prevalence of patient-reported swelling in the groin and leg increased fourfold. Swelling symptoms remained at 12 and 24 months after surgery, and seemed to profoundly affect perceived physical health and quality of life. [16]

Although the frequency of groin and leg swelling reported by the staff was 5%, 14% of patients reported swelling 3 months after the surgery. [16]

The diagnosis is clinical and remains too obvious in the presence of a large volume of the bursa, the scrotal skin becomes thick, cardboard, and loses its elasticity [17, 18]. Lymphatic edema can extend to the penis, causing sometimes urinary problems and preventing sexual intercourse [4, 19].

Paraclinical examinations contribute to the etiological diagnosis; Doppler ultrasound eliminates an obstacle on the vascular axis of the two lower extremities. In cases of filariasis, it shows adult worms moving within the lymphatic vessels. Ultrasound, abdominal computed tomography, and even MRI can rule out a compressive origin (pelvic or abdominal tumor) [20, 21]. Scrotal MRI can also be useful to specify the limits of surgical resection [22, 23].

While also considering the potential complications associated with elephantiasis, including papillomatosis, warty growth, lymph vesicles with embarrassing lymph oozing, and cellulitis. [5]

The treatment of penoscrotal elephantiasis is surgical and several techniques have been described. These can be grouped into two types of procedures:

Conservative technique or lymphangioplasty, aimed at improving lymphatic drainage using polyethylene or metallic tubes, omental grafts, or lymphatico-venous anastomosis. This involves anastomosing the saphenous arch to the superficial inguinal lymph node group. Unfortunately, the patency of this anastomosis is temporary [4, 24, 25, 26]. These techniques are currently being abandoned.

The second type of techniques includes radical excision of the elephantiasis tissue to prevent any recurrence. This excision allows for the removal of lymphatics, thereby performing a total superficial lymphangiectomy. Several plastic surgery techniques have been described for scrotal reconstruction after this radical excision. Among these techniques, we found pedicled skin flaps taken from the suprapubic or inguinal region [27, 4, 28, 30]. The thin free skin grafts can also be used [19, 26]. However, this method may alter local thermal regulation of the testes and induce disorders of spermatogenesis [4]. Another method can be used is the cranio-dorsal part of the scrotum, which is often preserved and allows for the reconstruction of a neo-scrotum. This method, utilized by many authors, appears to yield good functional and aesthetic results [4, 29,30, 19, 26, 18].

Regarding the penis, the use of thin free skin grafts (0.7 to 0.8 mm) yields the best results. This graft should be applied in a spiral manner to avoid longitudinal or circular retraction on the penis [27,4,29, 20, 31].

The results of this surgery are excellent with few recurrences. The persistence of a lymphatic blockage is implicated in the genesis of these recurrences [30, 31]

4. Conclusion:

Prostate cancer is the second most common cancer in men, and any complication arising after its therapeutic management should be of great importance. Penoscrotal elephantiasis is an obvious complication of its treatment, whether in the case of radical prostatectomy, with or without lymph node dissection (LND), or adjuvant radiotherapy if indicated. This condition has a negative impact on the function, mental state, and quality of life of patients already weakened by a neoplastic context. These findings suggest that it is important to explicitly inform patients about lymphatic swelling as a potentially persistent side effect, as well as the various therapeutic options, including the radical excision of the elephantiasis tissue followed by scrotal plasty, which allows for the reconstruction of a neo-scrotum using two often-preserved post-lateral scrotal flaps, providing excellent functional and aesthetic results with minimal recurrence.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional research committee and the 1964 Helsinki Declaration. Written informed consent was obtained for the publication of the photos.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

Author Contributions

H. EA contributed to the design and writing of the manuscript. A. D conceived and supervised the original project.

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