



Gigantic Suprapubic Lymphedema: A Case Study

Roozbeh Tanhaeivash¹, Tobias Franiel², Marc-Oliver Grimm¹, Marcus Horstmann¹

¹Department of Urology, ²Institute of Radiology, Friedrich Schiller University, Jena, Germany

We present the first case study of idiopathic gigantic suprapubic lymphedema and buried penis treated with puboscrotal reconstruction in a patient with initial extreme obesity after an extensive weight reduction (120 kg). Massive localized lymphedema of the suprapubic region should be differentiated from the scrotal type. Severe lymphedema could not resolve on its own and weight reduction does not seem to be helpful in such cases.

Key Words: Abdominoplasty; Adiposity; Elephantiasis; Reconstructive surgical procedures

Massive localized lymphedema (MLL), also known as pseudosarcoma, results from chronic lymphatic retention that can trigger chronic inflammation, fibrosis, tissue growth, and the development of angiosarcoma. Infections such as filariasis are the most common etiology in tropical countries, while obesity, trauma, radiotherapy, and lymphadenectomy are common etiologies in other parts of the world. MLL is most observed in parts of the body with excessive hanging soft tissue and relatively minimal lymphatic backflow, such as the arms, thighs, and abdomen [1,2].

In the male genitalia, this condition can result in buried penis with consequent difficulties in micturition and sexual dysfunction. Furthermore, it can lead to immobilization and problems regarding clothing. Although the first stage of treatment is weight reduction in extremely obese patients with buried penis [3], surgery appears to be more helpful in patients with persistent genital MLL.

We report the successful surgical treatment of an ex-

tremely obese patient in whom resection of MLL of the suprapubic region as well as puboscrotal reconstruction were performed.

CASE REPORT

We admitted a 52-year-old Caucasian man with a body mass index (BMI) of 38.3 kg/m² (weight, 120 kg; height, 177 cm) who had experienced a massive weight loss of 120 kg using diet alone (initial BMI, 76.6 kg/m²). His chief complaint was a gigantic genital mass extending beneath his knees in standing position that had progressed over the previous years (Fig. 1). At the time of admission, he was immobilized due to this giant mass, and it was impossible for him to wear normal clothing. Additionally, his penis was completely covered by the mass, so that micturition was only possible in the standing position with assistance. Therefore, a urinary catheter was placed. Most likely due to his mild bipolar disorder and schizophrenia, the giant

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Correspondence to: Marcus Horstmann

Department of Urology, Friedrich Schiller University, Lessing St. 1, 07743 Jena, Germany.
Tel: +49-2151-452-278, Fax: +49-2151-452-270, E-mail: marcushorstmann@gmx.ch

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genital mass had been neglected for a long period of time. As expected, the patient additionally reported impotence. The patient lived in a convalescent home and was single. He and his family were not able to take care of him at home due to his physical status and the gigantic case of lymphedema. Moreover, his bipolar disorder and schizophrenia occasionally caused difficulties for his caregivers, although these conditions were stable due to treatment with quetiapine and olanzapine at the time of admission.

The physical examination revealed a very large, painless, soft, and non-adherent tissue in the suprapubic region, completely covering the penis and scrotum. The skin was thickened and hyperkeratotic, but not erythematous. After moving the mass upwards, the penile orifice as well



Fig. 1. Ventral aspect of the massive localized lymphedema of the suprapubic region prior to surgery.



Fig. 2. The penile orifice and normal-sized testes can be seen after the mass is moved upwards.

as normal-sized testes could be observed (Fig. 2). His extremities were not edematous and no lymphadenopathy was detected. His medical history was otherwise unremarkable, apart from his stable bipolar disorder and schizophrenia. No history of trauma, radiotherapy, or tropical vacations was reported. In an abdominopelvic computed tomography scan, the huge genital mass demonstrated no signs of malignancy or inguinal hernia (Fig. 3). All laboratory findings, including tumor markers of testicular cancer, were within normal limits (α -fetoprotein = 2.6 ng/mL, beta-human chorionic gonadotropin <1.2 mIU/mL, and lactate dehydrogenase = 2.59 IU/L).

In order to facilitate normal urination and physical activities, as well as restoring acceptable sexual appearance and function, we performed a reductoplasty of the gigantic suprapubic mass as well as a puboscrotal reconstruction with a horizontal ellipsoid excision of the mass, leaving the penis and scrotum intact. The dilated veins and lymphatic vessels were meticulously ligated. Both spermatic cords were identified and marked using vessel loops (Fig. 4). Finally, a mass of 11 kg (measuring 40×27×14 cm) was resected (Fig. 5).

The histopathological examination revealed abundant lymphatic tissue and vessels with no sign of filariasis or a malignancy. Postoperatively, the wound healed by first in-



Fig. 3. A preoperative coronal computed tomography image. Right-pointing thick arrow, an enlarged lymph node; left-pointing arrow, dilated veins; circles, edematous soft tissue; stars, thickened cutis vera.



Fig. 4. Surgical steps leading to the resection of the mass.



Fig. 5. Postoperative appearance after ellipsoid tissue resection and puboscrotal reconstruction.

tention, and the patient was discharged with satisfying functional and cosmetic results. The bladder catheter was removed one month after surgery, once the wound had completely healed. The family of the patient reported that he exhibited no dysfunctions involving micturition. However, the patient was not able to function sexually due to impotence. The treatment of impotence was postponed due to the patient's psychological status. In fact, the patient never attended postoperative follow-up examinations and refused any further medical interventions. His family was contacted and informed about the procedures necessary for planning further medical interventions if requested by the patient, which would most likely involve penile reconstructive surgery.

DISCUSSION

Obesity (BMI $> 30 \text{ kg/m}^2$) is known to be a major etiology of severe health problems, such as atherosclerosis, coronary artery disease, diabetes mellitus, musculoskeletal disorders, and several types of cancer. Furthermore, some studies have highlighted the impact of obesity on quality of life as well as on the incidence of mood disorders, impaired sexual function, and sleep apnea. For instance, Hammoud et al [4] investigated 89 patients with a BMI $\geq 35 \text{ kg/m}^2$ who underwent Roux-en-Y gastric bypass surgery. The patients were evaluated regarding the quality of their sexual life, their metabolic and hormonal status, and the presence and severity of sleep apnea. Adiposity was found to have a significant association with unenjoyable sex, reduced libido, difficulties in sexual performance, and discontinuation of sexual activity. Moreover, a negative correlation was found between BMI and testosterone levels. Jagielski et al [5] studied 263 patients with a BMI $\geq 40 \text{ kg/m}^2$ or a BMI $\geq 35 \text{ kg/m}^2$ and a comorbidity, evaluating their quality of life and mental health status, and found a significant negative impact of adiposity on quality of life, especially in male participants. Moreover, Duval et al [6] evaluated 100 patients with morbid obesity who were candidates for bariatric surgery. They found that morbid obesity had a strong impact on many aspects of quality of life, such as interpersonal relationships, sexual life, physical activities, emotional functions, and personal hygiene. Additionally, obesity is a well-known predisposing factor for impotence and diabetes mellitus, meaning that pre-

vention, screening, and early treatment play an important role in the physical and mental health of the population [7].

Furthermore, adiposity and lymphedema have been hypothesized to have a reciprocal influence. Although secondary lymphedema has been observed in obese patients who had experienced iatrogenic trauma to the lymphatic system, primary lymphedema has likewise been postulated to occur more frequently in obese individuals more frequently, although the pathophysiology is unknown. However, lymphedema has been suggested to facilitate the differentiation of adipocytes, resulting in the additional deposition of fat with a concomitant inflammatory response. Therefore, weight management, screening for lymphedema, and the treatment of lymphedema are important for preventing morbidity and mortality due to these conditions [8].

In two reviews of patients with MLL, Chopra et al [1] and Brewer and Singh [2] reported MLL of the thighs to be the most frequent manifestation of this condition. They reported also their own cases of suprapubic MLL and the surgical management thereof. Chopra et al [1] reported MLL of the lower abdomen and scrotum to be the second and third most frequent variants of this condition, respectively. They observed angiosarcoma in 13% of the patients included in their study; moreover, they reported an overall mortality rate of 9.2%, with 66% of the patients with angiosarcoma dying of that disease. This suggests that early surgical interventions should play a primary role in the management of lymphedema, instead of planning weight loss programs or drainage and compression therapy. Brewer and Singh [2] reported the incidence of angiosarcoma to be approximately 7%, with a 100% mortality rate. They concluded that prevention is the best way to avoid the complications of this condition, in light of increasing trends in obesity.

Our patient presented with a gigantic suprapubic MLL. This condition should be principally differentiated from scrotal MLL, as they can be distinguished by a simple clinical examination even if they may seem to be similar in terms of size and general features. In scrotal MLL, the penile orifice is found on the ventral aspect of the hanging mass, in suprapubic MLL it can be discovered behind the mass once the hanging mass is lifted up (Fig. 1, 2). Scrotal

MLL is known to occur after pelvic operations, trauma, and radiotherapy, and may also occur due to inflammations of various etiologies (e.g., filariasis) or renal insufficiency [1,2].

To our knowledge, this is the first report of a patient with suprapubic MLL persisting after extensive weight loss.

Regardless of the etiology, surgery is the treatment of choice in all patients with extremely large chronic genital lymphedema. This is especially true if the mass is so large that it not only impairs micturition and sexual function, but also ambulation and the ability to wear normal clothing. However, in patients with mild to moderate lymphedema, conservative management (e.g., weight loss and physical activities, compression, and drainage) can be considered to be the first line of therapy, concomitantly with treatment of the underlying disorder (e.g., coronary artery disease, hypertension, or diabetes mellitus) [9]. Weight reduction, however, seems to have a paradoxical effect on lymphedema. On one hand, we would expect a notable weight reduction to lead to a regression in lymphedema due to a decrease in the pressure resulting from the mass of the body. On the other hand, some reports have demonstrated persistent lymphedema despite massive weight reductions [10]. Therefore, we suggest that weight reduction should first be attempted, as it has other benefits in patients with coronary artery disease, hypertension, and diabetes mellitus. Moreover, weight reduction may contribute to the resolution of lymphedema. If not, a surgical approach should be taken in order to prevent angiosarcoma [1,2].

Most importantly, the surgical approach should be carefully chosen and tailored to the clinical findings of the patient. In cases of suprapubic MLL, as occurred in our patient, the excessive tissue can be resected by a horizontal ellipsoid abdominoplasty, unburying both the penis and scrotum. However, this would be more difficult in cases of scrotal MLL, in which a vertical or T-shaped incision is first necessary to release the buried penis from the surrounding scrotum. If the penis is secured, the scrotum can be partially resected and most often must be reconstructed using the remaining non-scrotal tissue. Moreover, it is often not possible to preserve the penile skin, so that a penile mesh graft becomes necessary [9]. This, of course, makes such an intervention more difficult.

Before performing surgery to treat genital lymphedema (either suprapubic or scrotal MLL), extremely large scrotal hernias, filariasis, and malignancies should be ruled out.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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