

## Case Report

## Surgical Management of Giant Scrotal Lymphedema – A Case Report

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## Abstract |

Giant scrotal lymphedema (GSL) causes marked physical distress, and has severe cosmetic and psychosocioeconomic consequences, decreasing the quality of life of the sufferer. Excision is often required as medical treatment is insufficient to produce acceptable results. We report surgical management of a case of idiopathic giant scrotal lymphedema. Involved tissue was excised maximally. There was a significant improvement in walking, sitting and squatting, and patient was able to perform his daily activities in a much better way. At one year follow-up there was recurrence, for which further debulking was performed. Patient was satisfied with cosmetic outlook and his physical as well as psychological distress was significantly decreased.

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## Introduction

Giant scrotal lymphedema (GSL), also called scrotal elephantiasis is a rare condition characterized by massive scrotal enlargement with genital deformation. GSL is caused by inadequate lymphatic drainage due to aplasia, hypoplasia or obstruction of lymphatic vessels. This leads to accumulation of interstitial fluid in scrotal subcutaneous tissue. GSL not only causes physical distress but also has cosmetic and severe psychosocioeconomic consequences, decreasing the self esteem and quality of life of the sufferer. Excision is often required as medical treatment is insufficient to produce acceptable results.<sup>1,2,3</sup>

We report successful surgical management of a case of idiopathic giant scrotal lymphedema with no history of filariasis, malignancy, surgery or radiation.

## Case Report

Forty-year-old unmarried male presented with progressively increasing swelling of scrotum, right lower limb and left thigh for the past 20 years. Swelling was associated with itching, difficulties in maintaining

hygiene and recurrent skin infections with purulent discharge from the involved areas. Massive scrotal enlargement also caused heaviness, pain, difficulty in walking, sitting, squatting and urinating. Patient underwent drainage of recurrent abscesses multiple times. He had severe psychological distress due to inability to perform activities of daily living, foul odour and heavy expense of treatment.

Physical examination showed a massively enlarged scrotum extending down to knee-level. Penis was not visible and completely buried in the lymphedematous scrotal tissue. The scrotal skin was very thick, non-pitting and rugated, with multiple excoriations, pits and scar marks visible. It was not possible to palpate testes through the tough scrotal skin. The scrotal mass measured 30×32×34 cm (Figure 1 A, B & C). Inguinal lymph nodes were not palpable. Patient's gait was also disturbed.

Laboratory investigations were within normal limits and blood smear for microfilariae was negative. Abdominal and scrotal sonography revealed bilateral atrophic

testes. The patient was counseled and informed consent was taken before proceeding with surgery.



**Figure 1.** A,B& C: pre-operative appearance of scrotal mass; D & E: excised specimen; F: post excision wound with visible testes; G, H & I: immediate post-operative appearance



**Figure 2:** Same patient at 1 year follow-up showing recurrence of swelling (A & B), and after second debulking (C & D)

Surgical debulking of lymphomatous scrotal tissue was performed under general anesthesia. Patient was placed in lithotomy position and was catheterized. Skin incisions were marked and tumescent solution (1% lidocaine and 1:1000,000 epinephrine) was infiltrated. Involved tissue was excised maximally. Penis was

exposed all around and penile skin was found to be normal. Contrary to ultrasound finding both testes were also found normal in shape and texture (Figure 1F). Testes were repositioned and neo-scrotum was created with remaining spared posterolateral skin over a suction drain (Figure 1 G, H & I). Excised specimen weighing 16kg was sent for histopathology (Figure 1 D & E). Catheter and suction drain was removed on 7<sup>th</sup> post-operative day. The patient was discharged with scrotal support and also advised to wear tight compression garments.

Histopathology report showed nonspecific chronic inflammation with dilated lymphatic vessels, epidermal thickening and dermal fibrosis.

No postoperative hematoma, infection, necrosis or wound dehiscence was observed. Pain and dull aches were markedly reduced. There was a significant improvement in walking, sitting and squatting and he was able to perform his work in much better way. At one year follow-up there was recurrence, for which further debulking was performed (Figure 2).

### Discussion:

We found that surgical debulking for giant scrotal lymphoedema (GSL) is very rewarding. The patient reported marked psychological relief, and was satisfied with functional and cosmetic outcome. The procedure improved his quality of life.

Genital lymphedema is a rare condition and is classified as congenital (primary) and acquired (secondary). Primary lymphoedema is further classified into congenital hereditary lymphoedema, lymphoedemaprecox and lymphoedema tarda based on the age of onset in infancy, childhood and adulthood respectively. Most common cause of Secondary lymphoedema is filariasis, however it can also result secondary to radiotherapy, surgery, malignancy or trauma.<sup>3,4,5</sup>

Shusruta was thought to be the first surgeon who performed excision in genital lymphedema at around 600 BC.<sup>3</sup> Some surgeons reported serial debulking of lymphomatous scrotal subcutaneous tissue, however other surgeons preferred near total excision of abnormal tissue.<sup>2,3</sup> Similarly different surgical techniques were described for coverage of exposed testicles after resection including skin grafting or reconstruction of neo-scrotum with anterior, posterior or posterolaterally based remaining scrotal skinflaps or regional pedicled flaps from thigh.<sup>6</sup> Historically some surgeons reported burying testis in pouch made in thigh. Jones et al found

that higher temperature resulting from pedicled flaps from thigh for neoscrotum reconstruction led to disturbed spermatogenesis and used remaining posterior scrotal skin for coverage of the exposed testicles.<sup>6,7,8</sup>

We strengthen that excision is mainstay of treatment of GSL. In this case our aim was to decrease the physical distress of the patient, so maximum debulking was performed with preservation of testis and the cord. As described in few other studies, studies we also used posterolateral skin for neoscrotal reconstruction as it was relatively spared.<sup>8,9</sup> The purpose of making neoscrotum with posterolateral skin was to provide natural tissue for cover, which will maintain normal testicular function and also provide better cosmesis. Minor complications such as wound infection, dehiscence and delayed wound healing were mentioned in literature however in this case we did not encounter any complication.<sup>12</sup> We observed poor postoperative patient compliance for non-surgical adjunctive measures (massage, elevation and compression garments) and that might be a reason for recurrence seen at one year follow-up.

Our Patient was satisfied with cosmetic outlook and his physical as well as psychological distress was significantly reduced. He no longer has recurrent scrotal skin infections. He was able to return to work and has become a useful member of his family. He is also planning for marriage.

In our opinion properly planned surgical debulking should be performed for this incapacitating condition and making neoscrotum with relatively spared posterolateral skin maintains testicular function with pleasing cosmesis.

## References

1. Mitre AI, da Silva JCF, Cintra W, Quagliano AP, Arap S, Ferreira MC. Surgical treatment of lymphedema
2. Modolin M, Mitre AI, Silva JC, Cintra W, Quagliano AP, Arap S, Ferreira MC. Surgical treatment of lymphedema of the penis and scrotum. *Clinics*. 2006 Aug;61(4):289-94.
3. Hodjati H, Khezri AA. A new modified surgical approach in lymphedema of the penis and scrotum. *Med J of The Islamic Republic of Iran*. 1996 Aug;10(2):113-5.
4. Brotherhood HL, Metcalfe M, Goldenberg L, Pommerville P, Bowman C, Naysmith D. A surgical challenge: Idiopathic scrotal elephantiasis. *Canadian Urological Association Journal*. 2014 Jul;8(7-8):E500
5. Dianzani C, Gaspardini F, Persichetti P, Brunetti B, Pizzuti A, Margiotti K, Degener AM. Giant scrotal elephantiasis: an idiopathic case. *International journal of immunopathology and pharmacology*. 2010 Jan;23(1):369-72.
6. de Souza Filho MV, de Almeida Girão R, Tomás Filho ME. The challenge of surgical treatment for giant scrotal lymphoedema-the role of local flaps. *Journal of Plastic, Reconstructive & Aesthetic Surgery*. 2010 Feb 1;63(2):e187-8.
7. Ollapallil JJ, Watters DA. Surgical management of elephantiasis of male genitalia. *British journal of urology*. 1995 Aug 1;76(2):213-5.
8. Jones HW, Khan RA: Surgical treatment of elephantiasis of the male genitalia. *Plast Reconstr Surg* 46: 8, 1979.
9. Garaffa G, Christopher N, Ralph DJ. The management of genital lymphoedema. *BJU int*. 2008 Aug; 102(4):480-4.