Giant scrotal lymphoedema – A case report
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ABSTRACT

INTRODUCTION: Giant Scrotal Lymphoedema is a rare disease. Such Scrotal elephantiasis presents multiple problems both to the patient as well as the treating clinician obstruction, aplasia, or hypoplasia of the lymphatic vessels. The most common cause world wide is lymphatic Filariasis.

PRESENTATION OF CASE: We present a particularly grotesque where the resected scrotal tissue weighed 32 kg which is one of the largest so far mentioned in literature. The lymphoedema was progressive over 8 years duration and the testes were not palpable with the penis deeply buried.

DISCUSSION: Scrotum was explored and penis was recovered deep within the pit of lymphoedema. Careful dissection done with cautery to delineate penis circumferentially from the root of scrotal lymphoedema. Foleys catheterisation was done. After the separation of penis scrotal skin flaps were raised on either side by extending the incision horizontally. De bulking of lymphoedema was done and the remaining scrotal skin was closed in Y shaped manner with root of penis in centre. Meticulous technique of dissection, cautery and ligasure use of Ligasure enabled excision with minimal blood loss.

CONCLUSION: Once fibrosis sets in resectional therapy will be needed in most cases. Successful reduction scrotoplasty with acceptable cosmetic results can be obtained in giant scrotal lymphoedemas weighing as large as 32 kg as in our case.

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1. Introduction

Giant Scrotal lymphoedema is rare even in region endemic for filariasis. Recently McDougal presented classification 1 of the causes of lymphoedema of the external genitalia. Massive scrotal lymphoedema sometimes called Scrotal elephantiasis causes immense physical as well as emotional distress to the patients. Scrotal elephantiasis, or massive scrotal lymphoedema, is a disease caused by obstruction, aplasia, or hypoplasia of the lymphatic vessels draining the scrotum. It can be either congenital or acquired in nature, with the most common acquired etiology being infection, mainly lymphogranuloma venereum or filarial infestation with Wucheria bancrofti. While the etiology of lymphoedema does influence the management strategy, once fibrosis sets in resectional therapy will be needed in many cases. 2

2. Presentation of case

A 54 years old male patient, with a weight of 110 kg was admitted with bilateral massively enlarged scrotum. The scrotal lymphoedema had started 8 years earlier and had been relentlessly progressing to reach the presenting size of enormous proportion. There was no history of any radiation. A previous unsuccessful attempt of partial resection was done at an early stage of the scrotal enlargement and, after this procedure, the patient refused to be reviewed by physicians because he felt embarrassed. In fact he could not even walk easily and used to sleep on the floor because was not able to get on the bed (Fig. 1).

Clinical examination revealed giant elephantiasis totally engulfing the penis. Urine was voided from deep pit on the scrotal skin. However scrotal skin was reasonably healthy. Testes were not palpable. All the routine blood investigations were found normal. Imaging studies revealed bilateral atrophic testis. Neither clinical examination nor investigations revealed any other abnormality in the abdomen, inguinal regions or in the lower limbs. Though the cause of the lymphoedema was thought to be filariasis owing to its endemicity the same could not be proved by investigations. After considering the size of the lesion, patient's age and social circumstances decision was taken to do near total scrotal skin and soft tissue excision. As the penis was buried deep, the pit in the scrotum was explored. Careful dissection done with cautery to delineate penis circumferentially from the root of scrotal lymphoedema. Foleys catheterisation was done. After the separation of penis scrotal skin flaps were raised on either side by extending the incision horizontally. Large blood vessels (few mms to 3 cm) were tied, tran fixed and cut with ligasure. In view of the atrophic nature of the bilateral testis and the fact that they were located deep within the mass of tissue necessitating extensive dissection to reach them it was decided to do bilateral orchidectomy and leave the cords behind. It was postulated that the cords might have formed an alternate pathway for lymphatic drainage. De bulking of lymphoedema was done and the remaining scrotal skin was closed in

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Y shaped manner with root of penis in centre. Meticulous technique and use of Ligasure enabled complete excision with minimal blood loss. The specimen measured and weighed 32 kg (Fig. 2a). The duration of the procedure was 3 h and 20 min. The postoperative period was uneventful and sutures were removed after 2 weeks. Minor wound infection was controlled with antibiotics and regular dressings. Patient was discharged with advice to wear tight undergarment.

The histopathology revealed extensive fibrosis of the tissue along with lymphostasis. The testis was confirmed to be atrophic.

3. Discussion

Giant scrotal edema is a rare entity even in areas endemic for filariasis. Mc Dougal classified lymphoedema of external genitalia in to congenital and acquired. Filaria is the most common cause of acquired genital edema world wide. Radiation, neoplasms, granulomatous diseases are the other causes of genital lymphoedema. In lymphatic filariasis the main lymphatic channels become dilated and flow is impaired. The network of peripheral lymphatics in the skin becomes a ‘safety valve’ and acts as a conduit through the skin until healthy lymphatic trunks are found proximally to the damaged vessels. Lymphostasis and lymphotension lead to the accumulation of interstitial fluid, proteins growth factors and other active peptide moieties, glycosaminoglycans and particulate matter including bacteria. As a consequence there is increase in collagen production by fibroblast, an accumulation of inflammatory cells (predominantly macrophages and lymphocytes) and activation of keratinocytes. The end result is protein rich edema fluid, increased deposition of ground substance, subdermal fibrosis and dermal thickening and proliferation.

Clinically the swelling is painless and shows pitting edema atleast initially. Gradually the skin becomes coarse and tough as fibrosis sets in such massive enlargement of genital skin may cause multiple symptoms like disfigurement, urinary dribbling, impotence, recurrent cellulitis. Apart from the physical infirmity the emotional disturbance is a major factor in overall health. Restoration of lymphatic drainage by lymphangioplasty has been attempted with limited success in minor cases without fibrosis. In more advanced cases excision of all the affected skin and soft tissue forms the mainstay of the treatment. The procedure is sometimes called reduction scrotoplasty. Posterior skin of the scrotum can be used to reconstruct the scrotum as it is generally less diseased. The possible operative complications include haemorrhage, damage to the urethra, hematoma, wound infection and of course recurrence of the lymphoedema.

4. Conclusion

Giant Scrotal lymphoedema is a distressing condition causing both physical and psychological distress to the patient. Excisional surgery with reconstruction is the mainstay of the treatment. In our case near total excision of scrotum with reconstruction was done (Fig. 2) with acceptable cosmetic result.

Conflict of interest statement

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Consent

Written consent has been obtained from the patient.

Author contributions

All authors contributed equally.

References