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Trauma and reconstruction

Giant scrotal lymphedema after paraffinoma injection: A case report

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ABSTRACT

Scrotal lymphedema is a rare condition characterized by blocked lymphatic drainage to the scrotum, resulting in physical and psychological discomfort for sufferers. Here, we present a case study of a 27-year-old male with giant scrotal lymphedema resulting from paraffinoma injection. The patient experienced scrotal enlargement since 2019, which engulfed the penis and was surrounded by edema. After confirming the absence of filariasis parasites, the patient underwent paraffinoma excision and scrotoplasty, resulting in a 13 kilograms scrotal specimen with no signs of malignancy. Giant scrotal lymphedema can be distressing, but surgical excision can improve the patient's quality of life.

1. Introduction

Scrotal lymphedema is a rare condition in which the lymphatic system draining to the scrotum becomes blocked. Filariasis is the most prevalent cause of scrotal lymphedema, but it can also be caused by radiation, neoplasms, or granulomatous disorders. Limitation in physical activities as well as psychological discomfort in sufferers are two considerable symptoms when the disease becomes large enough. ^{1,2} We present a case of giant scrotal lymphedema that arises after paraffinoma injection.

2. Case presentation

A 27 year-old male was referred from Hospital in Ambon, Maluku with chief complaint scrotal enlargement since 2019. Patient had a history of silicone injection to the penis in 2017 and started feeling discomfort and difficulty to walk since 2019 because of the scrotal enlargement. Scrotal tenderness and fever were denied. Physical examination showed normal vital signs with local examination revealed scrotal enlargement totally engulfing the penis surrounded by edema (Fig. 1a–c). Testes were not palpable. Transillumination test was positive, there's no fluctuation and erythema. The laboratory showed slight leukocytosis (14.400) and slightly elevated liver enzyme. Non-contrast whole abdomen computed tomography scan (CT scan) showed liver enlargement, massive edema in scrotum bilateral clinically consistent as massive lymphedema. Patient was from the endemic place of filariasis

(Ambon) and was checked for peripheral blood smear, the result showed no sign of filariasis parasites.

Patient then treated with paraffinoma excision and scrotoplasty by identifying funiculus spermaticus, excision of the scrotum and scrotoplasty, shaft of penis felt tender and excision of paraffinoma was done in the shaft penis. Drain was placed, the raw surface was closed (Fig. 2a and 2b). Specimen of scrotum weighed 13 kilograms with size $53\times42\times26$ cm (Fig. 2c) and tissues were sent to pathology. Patient was hospitalized and discharged four days after the procedure with minimal pain and no sign of infection (Fig. 3a–e). Results from pathology revealed angiomyxoma and granulomatous inflammatory reaction without signs of malignancy.

This is a very rare case where we had the largest scrotal enlargement we ever recorded due to paraffinoma injection. However the outcome was great and the patient felt very satisfied. The patient came several times to our polyclinic on some appointments for follow up, and the patient did not have any complaints or adverse events resulting from the procedure.

3. Discussion

Lymphedema is an abnormal buildup of protein-rich fluid in soft tissues caused by a malfunction of the lymphatic drainage system. There is an imbalance between lymph production and absorption, thus leading to fluid build up in the area and leading to swelling and discomfort. In chronic circumstances, the condition manifests as fat accumulation and

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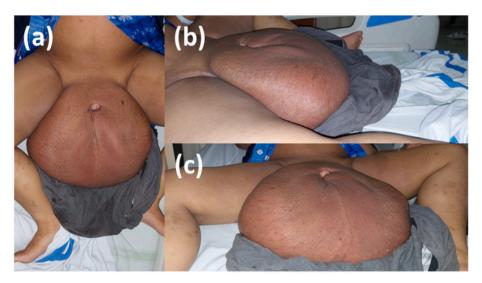


Fig. 1. (a). Giant scrotal lymphedema clinical appearance, (b). Patient's penis was not visible from lateral view, (c). Scrotal lymphedema pictured from below.

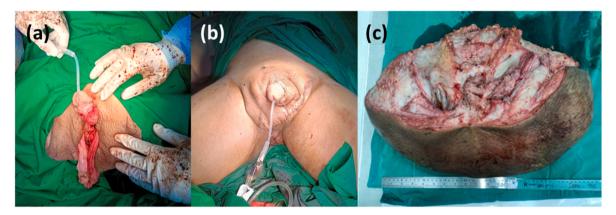


Fig. 2. (a). Clinical appearance after paraffinoma excision, (b). Scrotal appearance after scrotoplasty procedure, (c). Scrotal specimen that was sized $53x42 \times 26$ cm and weighed 13 kilograms.

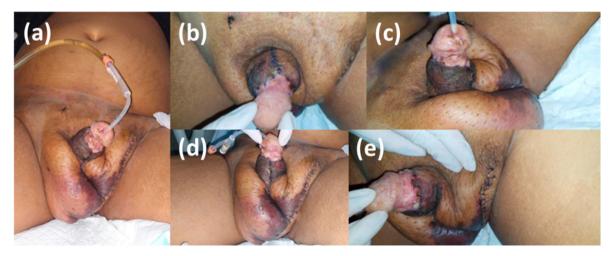


Fig. 3. (a). Post-operative paraffinoma excision and scrotoplasty day 4, (b). Clinical apperance of penile after surgery from top (c). Right lateral view of scrotum, (d). Bottom view of scrotum, (e). Left lateral view of scrotum.

fibrous tissue thickening. The restriction of lymphatic flow produces ductal dilatation, as well as connective tissue hypertrophy and hyperplasia, interstitial edema, and chronic inflammation.²

It is estimated that there are approximately 200 million cases of

lymphedema worldwide.³ It can be congenital or acquired, with the most frequent acquired cause being infection, namely lymphogranuloma venereum or filarial infestation. This kind of tropical disease usually occurred due to filarial parasites transmission through mosquito

bite.4

Patients mostly come because of limitations in physical activity related to the scrotal enlargement. The symptoms of scrotal lymphedema can range from mild to severe and may include a feeling of heaviness or fullness in the scrotum, discomfort or pain, and difficulty moving the affected area. As the patient came from an endemic location of filariasis (Ambon) and had a previous history of silicone injection, peripheral blood smear should be done to exclude the cause and confirmed with CT-Scan to exclude other diagnosis including tumor.

Silicone injections to the penile were common in some places in Indonesia and the complication of the procedures were differ on each case including scrotal enlargement from silicone injection or penile augmentation through paraffin injection. The case usually occurs due to the mechanism of moving silicone substances that leaks out of the ruptured implant and thus can cause scrotal lymphedema. Moreover, the increased volume of injection material used in scrotal augmentation raises the likelihood of granuloma formation. Another cause of scrotal lymphedema is paraffin injection. A penile enlargement method that is usually performed by a non-professional health worker, by performing paraffin injections to the penile.

Due to its symptoms, giant scrotal lymphedema can be quite distressing both socially and physically. While the cause of lymphedema influences care, once fibrosis develops, resectional therapy is usually required. ^{2,5} Furthermore, treating these instances is frequently difficult for surgeons. Surgical excision, on the other hand, can be a viable alternative. Removing the substance and reconstructing the affected area through surgery is the main method of treating penoscrotal edema, along with scrotal preservation. Indocyanine green administration is usually performed to aid precise lymph node identification. However we did not consider this option because the patient had a history of multiple drug allergies.

In general, all patients who underwent surgical therapy experienced significant improvements in their quality of life, penile feeling, and sexual intercourse. $^{2.5}$ In addition, the patient experienced minimal pain and was delighted with the outcome, and improved appearance within a 2–3 year period after the surgery. In our situation, surgical excision was unquestionably required because histopathological investigation revealed angiomyxoma and granulomatous inflammatory reaction.

4. Conclusion

Scrotal lymphedema is a rare condition characterized by scrotal enlargement caused by a breakdown in lymphatic outflow. A 27-year-old male referred from endemic location of filariasis with scrotal enlargement and history of silicone injection to the penis 2 years before enlargement and was treated with paraffinoma excision and scrotoplasty. Patient was discharged with minimal pain and delighted with the outcome. While establishing diagnosis and cause of the enlargement can be challenging. Treating patients with surgical excision can improve the quality of life of the patient.

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Declaration of competing interest

Author declares no competing interest in creation of this paper.

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References

- Vives F, García-Perdomo HA, Ocampo-Flórez GM. Giant lymphedema of the penis and scrotum: a case report. Autopsy & Case Reports. 2016;6(1):57.
- Binjawhar A, El-Tholoth HS, Alzayed EM, Althobity A. Successful surgical treatment
 of giant scrotal lymphedema associated with Hodgkin's lymphoma: a rare case report.
 Urology Case Reports. 2021;37, 101708.
- Greene AK, Slavin SA, Brorson H. Lymphedema: Presentation, Diagnosis, and Treatment. Springer; 2015.
- Irdam GA, Fadhly S. Scrotal reconstruction on scrotal lymphedema. Urology Case Reports. 2018;20:9.
- Ringoringo DRL, Nasution R, Adhyatma KP. A single-stage reconstruction on giant scrotal lymphedema: a case report. Bali Med J. 2022;11(1):387–390.