Case Report

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Giant Scrotal Lymphedema: A Presentation of Rare Urogenital Disease – A Case Report

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Abstract

We report a case of 21-year-old man with scrotal lymphedema which weight was 36 kg. The patient has been undergone surgery where we completely removed the affected tissue, after this intervention we achieved good outcomes and increased the quality of patient's life. Giant Scrotal lymphedema is ill health which inflict physical and psychological damage on the patient. So total surgical excision the affected tissue with reconstruction is a principal method of advanced scrotal lymphedema.

Keywords: Scrotum lymphedema; Aplasia; Hypoplasia; Filariasis

Introduction

Giant Scrotal lymphedema is rare urogenital disease. The aplasia, hypoplasia, obstruction or any pathological changes of the scrotum lymphatic vessel led to occur the lymphedema. As a sequence of this process, surplus fluid piles up in the scrotum and we can revealing increasing of genitalia's size, thickening and hardening of the skin. Lymphedema divided into 2 types: a primary and secondary. Primary lymphedema is caused by disturbing of normal develop lymphatic vessels due to genetic mutation [1-4]. Secondary lymphedema occur after influence of trigger factors which lead to damage lymphatic vessels. The purpose of this article is to describe the first experience of treatment of giant scrotal lymphedema in Uzbekistan, where the resected scrotal tissue weighed 36 kg which is one of the largest so far mentioned in literature.

Case Report

A 21-year-male patient, was admitted with bilateral massively enlarged scrotum and was treated from 24-04-2018 to 14-06-2018. He complained on increase scrotum, difficult urination, difficult walking and sleeping, lack of sex, swelling of left lower limb, general weakness He has been suffering from this disease since childhood. The scrotal lymphedema started in 2010 year and was relentlessly progressing to reach the presenting size of enormous proportion **(Figure 1)**. From 6-10-17 to 17-11-2017 patient had been taking diagnostic examinations in our centre.

His general condition was good, and he had normal vital parameters. On examination, the patient had a massively enlarged scrotum extending below his knees. The huge solid verrucous scrotal mass of 80 × 90 cm, edema of scrotal skin and close tissues made it impossible to differentiate the external genitalias and the urethral orifice emerged as a deep pit on the anterior surface of the mass. The scrotal skin was thickened and edematous hiding the penis. There was swelling of the left lower extremities and edema of left arm. The testicles and cords were not palpable.

Laboratory tests including human immunodeficiency virus, markers for testicular cancer, and antibodies to schistosomes, chlamydia trachomatis and filariae were all negative. An ultrasound scan of the abdomen showed hepatic haemangiomas with less than 2 cm in diameter in the 7-8 segments. A CT scan indicated enlarged axillary, retroperitoneal, lymph nodes up to 1.3 cm. Mediastinal, bronchopulmonary lymph nodes were not changed. Massive edema of the subcutaneous fat at the level of the abdominal wall and pelvis-swelling extends into the tissue of the walls of the chest. There is formation with an inhomogeneous structure; size was 42.0 × 22.0 × 36.0 sm at the level of the perineum and the scrotum. This formation came out from the scrotum. Massive edema of subcutaneous tissue on the minor pelvis and left lower extremities. Iliac lymph nodes were up to 1.2 cm, inguinal lymph nodes on the background of edematous tissue are not differentiated separately **(Figure 2)**.

We decided to perform a subtotal scrotectomy with preservation of the penis and both testicles and subsequent reconstruction of the scrotum using locally tissues. The first step of the surgical procedure was the mobilization of testis. On the right side parallel to the inguinal crease we did 15 cm skin incision. During mobilization, a lot of lymphatic fluid was released from the tissue.

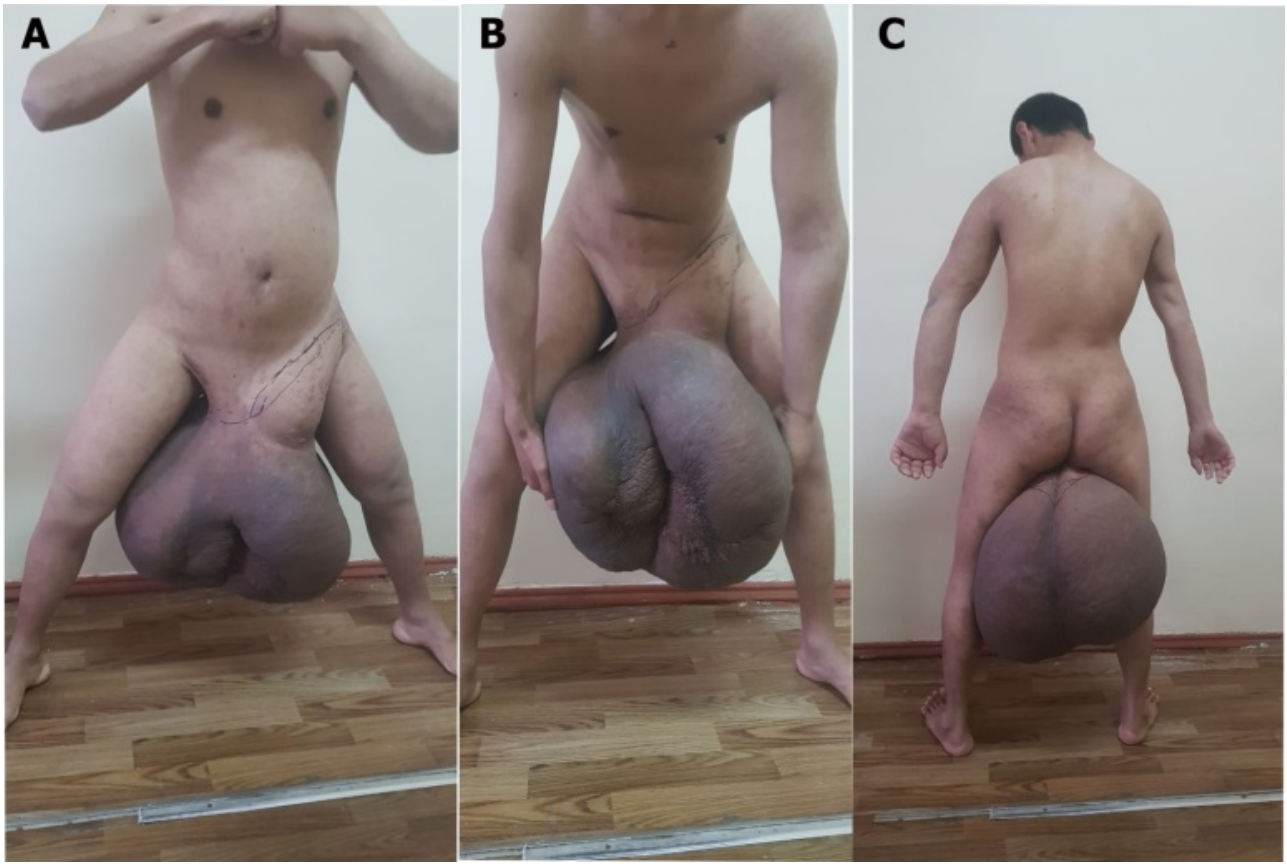

Figure 1 Giant scrotal lymphedema (A and B-Anterior view and C-Posterior view).

Figure 2 CT scan of the abdomen and pelvis.

Subcutaneous adipose tissue was thickened to 10 cm. Aponeurosis was dissected and spermatic cord was discovered, which was thickened and 30 cm long. When we palpated the testicles and spermatic cord they were not changed. Oriented on direction of the spermatic cord we isolated testicle. We weren't able to pull the testicle out of because of adhesion process and size of testicle. So we decided to make incision in the scrotum. After opening own testicular membrane we aspirated 2.0 litres of yellow-coloured liquid from the both testicles. All layers of testicles were stretched and thinned. Bergmann's technique was used for the surgery on testicles.

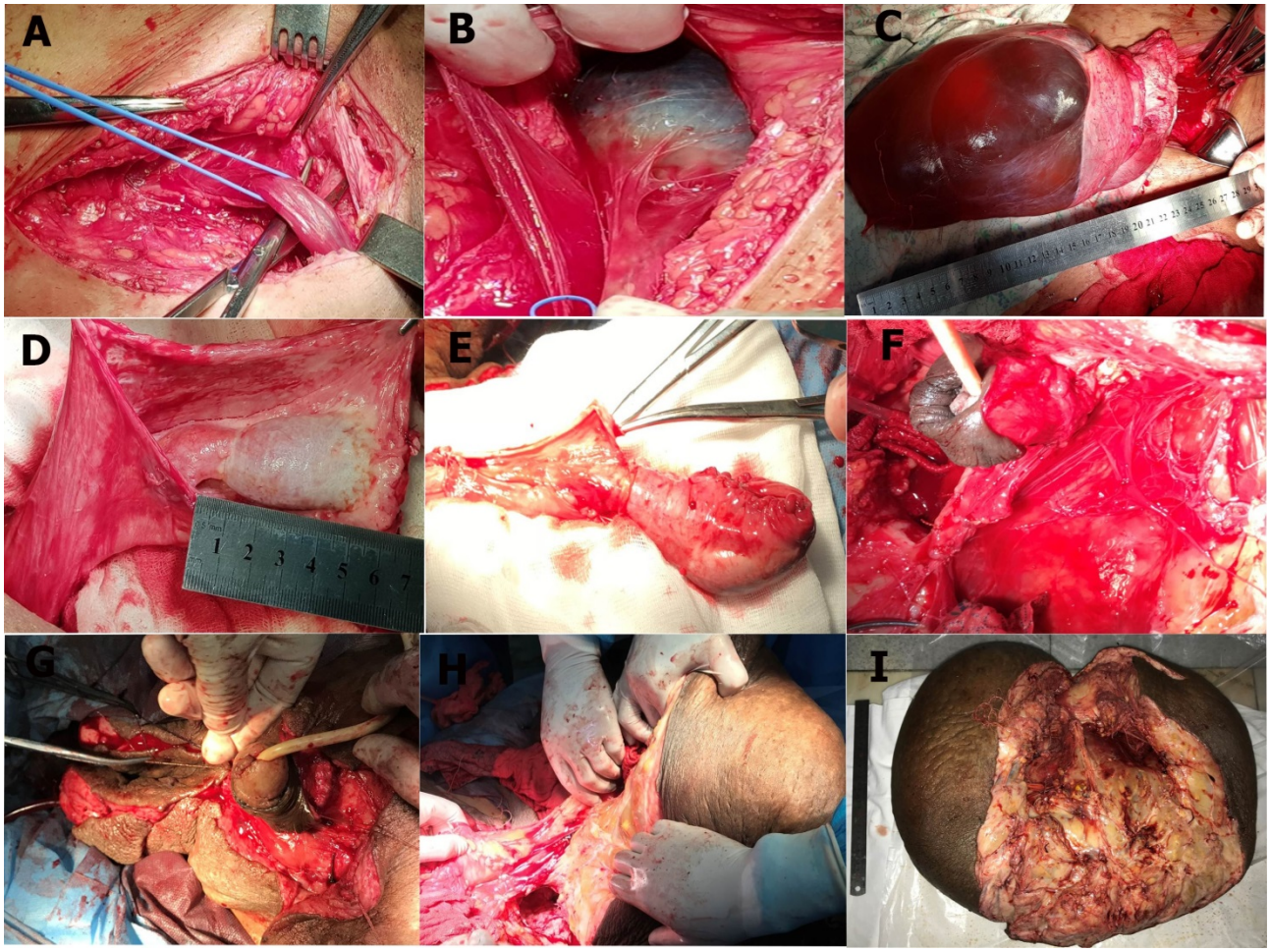

Figure 3: A- Mobilization of the spermatic cord; B- Mobilization of the right testicle; C- Hydrocele of the right testicle; D- Own testicular membrane after opening; E- Bergmann's technique; F- Mobilization of penile "tunnel"; G- Plastic penile; H- Dissection of the scrotum; I-The resected scrotum weighing 36 kg.

Figure 4 A- Appearance after operation; B, C- Appearance of the scrotum 15 days postoperatively; D- After 3 months.

The next step of surgery was mobilization of penile "tunnel" and the penile shaft was uncovered. A Foley catheter was placed. The involved skin of the penile shaft and prepuce was excised. The huge mass of the scrotal tissue was excised. Both of the testicles were put into artificial scrotum, which we made from surrounding tissues and drained. The defect of skin was reconstructed from the surrounding local tissue **(Figure 3)**.

During surgery, approximately 2.5 litres of blood were lost. The size of removed scrotum was 80 × 70 cm, weight was 36 kg. The histopathology revealed cystic extended lymphostasis of the scrotum. At postoperative period patient was treated with antibiotics and transfused with plasma and blood products. The post-operative period was uneventful and sutures were removed after 2 weeks, drainages were removed

after 7 days **(Figure 4)**. Patient was also treated by psychotherapist. The sexual function of the reproductive organs is not impaired.

Discussion

Scrotal elephantiasis is a rare disease. Depending on etiology factor of lymphedema it is divided into 2 types: a primary and secondary. Most common causes of the second lymphedema are infection, neoplasma, trauma or chronic venous conduce to thrombosis and fibrosis of the lymphatic channels. The filarias, nematode infection endemic to regions of South America, Asia and Africa are the main cause of secondary lymphedema in the world. In some dermatological disease like Hidradenitis suppurativa and lastly Angioneurotic lymphedema can develop phlebitis and lead to lymphedema [1-3]. Primary lymphedema has heterogeneous symptoms and different manifestation times and it is subdivided into forms: congenital lymphedema, lymphedema praecox, and lymphedema tarda [4]. Primary lymphedema unlike secondary lymphedema most common involves the neck, conjunctiva, and genitalia with any limbs included [5]. It is also can be hereditary disease [6].

The diagnosis of lymphedema is made clinically. Also important the family history for establishing of primary lymphedema and confirmed by fluorescence imagining (NIFR) or lymphoscintigraphy test [7]. Ultrasonography, computed tomography and magnetic resonance imaging help to assess the existence of extra fluid in other organ.

The management depends on accuracy of diagnosis, successful establishing stage of disease. There are 2 options for treatment in lymphedema: non-surgical and surgical. The disease which are underlying of lymphedema must be treated. Especially for treatment veneral disease must use antibiotic [8], for inflammatory diseases-steroid therapy [9,10], electrolytic imbalances must be normalized and parasitic infections treated by anti-parasitic medications. Absence of cancer must be confirmed. Lymphedema of the genitalia is more spreading in developing countries [11] and 20% of the male population may be affected [12]. The lymphatic drainage pattern of the cord structures and the testes are independent therefore they are not involved into the disease [13] and their important reproductive, sexual and hormonal functions must be saved, whichever the type and scope of surgical intervention. In chronic, advanced cases complete removing of all the affected tissue preclude recurrence. In our case we have removed all affected tissue with a preservation of the penis, spermatic cord and testes. After 18 months of follow-up, the patient's erectile function was restored, allowing sexual intercourse.

Conclusion

Giant Scrotal lymphoedema is a distressing condition causing both physical and psychological distress to the patient. This case vindicates that surgical excision with reconstruction of the advanced scrotum lymphedema stage can be performed with satisfactory results.

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