

CASE STUDY

# Isolated Giant Scrotal Lymphoedema From Filariasis in a Nigerian Male: A Case Report

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## ABSTRACT

Isolated lymphoedema of the scrotum is rare. In sub-Saharan Africa, the infective cause is commonly due to lymphatic filariasis, a neglected tropical disease commonly caused by *Wuchereria bancrofti*. The swelling of the affected skin and soft tissues results from impaired lymphatic drainage of the affected area due to the chronic inflammatory response to the dead and dying filaria worms in the lymphatic vessels. We describe the case of a 38-year-old man who presented with isolated scrotal lymphoedema secondary to *W. bancrofti* infection. The patient, in addition to cyclical anti-filarial medical treatment, had successful surgical resection of redundant and fibrous scrotal tissue resulting in overall improvement in his quality of life.

**Keywords:** Filariasis, Lymphoedema, Scrotal.

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## 1. INTRODUCTION

Lymphatic filariasis is a mosquito-borne roundworm infestation caused by *Wuchereria bancrofti*, *Brugia malayi* and *Brugia timori* [1]. It is one of the neglected tropical diseases (NTD), and the leading causes of disability from infection worldwide [2]. Massive scrotal lymphoedema from filariasis (Scrotal elephantiasis) is caused by obstruction of the lymphatic channels draining the scrotum, a consequence of chronic inflammatory response to dead or dying filarial worms. In sub-Saharan Africa, *W. bancrofti* infestation remains the most common aetiology of lymphatic filariasis and could present as lymphoedema of the extremities, genital and breast [1], [3]. Isolated genital lymphoedema is a rare presentation that is associated with immense physical as well as emotional distress to the patient as the genital could be swollen to alarming proportions [3]. Lymphoedema of the penis and scrotum causes mobility and voiding limitations, fatigue, pain, and recurrent subcutaneous infections due to difficulty with self-hygiene. It also causes sexual limitations, social isolation, and impaired quality of life [4].

In 2020, 863 million people living in 50 countries were at risk of becoming infested with lymphatic filariasis [5]. The current global burden of the disease is such that 25 million

men are living with hydrocele, over 15 million people have lymphoedema and about 36 million people live with the chronic manifestation of the disease [5].

There are few reported cases of isolated scrotal lymphoedema from filarial worm infestation [3], [6]. The other commonly identified aetiology is Lymphogranuloma venereum [3]. While the aetiology of genital lymphoedema affects the management strategy, surgical resection of the redundant scrotal fibrous tissue, cyclical medical therapy, and psychosocial support are the mainstay of the management plan in filarial causes [3]. We present a case of isolated giant scrotal lymphoedema from filariasis.

## 2. CASE REPORT

The patient is a 38-year-old, single, riverine farmer, who presented to us with a 10-year history of progressively increasing scrotal swelling which was insidious in onset, but progressively increased over time to the current size with the fundus at the level of the knees. This was associated with scrotal itching and recurrent excoriation/s/ulcerations and consequent variegated loss of scrotal skin colour but there was no history of scrotal pain. There was no associated swelling of any other part of the body. There





Fig. 1. The scrotal mass with partial burying of the penis.

was no history suggestive of heart, kidney, or liver dysfunction. There was also no history of lower urinary tract symptoms, haematuria, penile discharge, chronic cough, prolonged lifting of heavy objects, weight loss, or retroviral disease. There was no preceding trauma to the external genitalia, urinary tract instrumentation, or irradiation of the pelvic region. History of excessive mosquito bites both at work and at home could not be gotten. His erection was poor and his ambulation was limited due to the illness. He was often in a low mood and had to suspend his tertiary education because of the illness. For the above complaints, he had presented to several traditional homes and hospitals with no relief. He was referred to our facility from a private healthcare facility for further management.

On examination, he was a healthy-looking young male that was afebrile, anicteric, and not pale with no pedal oedema. He had a massively enlarged, non-tender, non-fluctuant, scrotum measuring about 60 × 30 cm, with the fundus at about his knee level. The scrotal skin was thickened, furrowed, and non-pitting, with areas of cracks/crevices, hypo- and hyperpigmentation. The penis was almost buried but had no abnormal findings (see Fig. 1). Both testes were impalpable and there was no abnormal finding on digital rectal examination. The patient, however, had bilateral tender inguinal lymph nodes.

Peripheral blood film assay for microfilaria at 10 pm, 2 am, and 12 noon were positive for *Wuchereria bancrofti*. He had normal renal and liver function tests, non-reactive Venereal Disease Research Laboratory (VDRL) screening for syphilis, and negative serology for chlamydia infection. Viral screening for HIV-1 and 2, hepatitis B and C infections were non-reactive. Full blood count showed a normal white blood cell pattern and erythrocyte sedimentation rate of 52 mm/hour. Seminal fluid analysis showed azoospermia. Scrotal ultrasound showed bilateral inguinal lymphadenopathy with signs of chronic scrotal lymphoedema. The testes were 8.6 and 9.1 cm<sup>3</sup> on the right and left sides, respectively. There was no evidence of varicocele or hydrocele. Urine analysis, microscopy, and culture were essentially normal.

He was counseled on the diagnosis and management plan which was multidisciplinary (medical and surgical

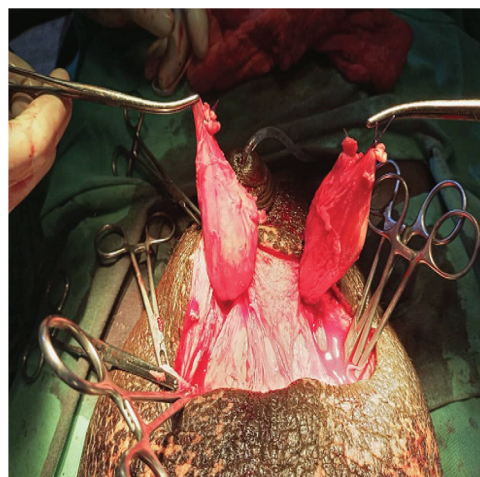


Fig. 2. Identification and isolation of the testes.

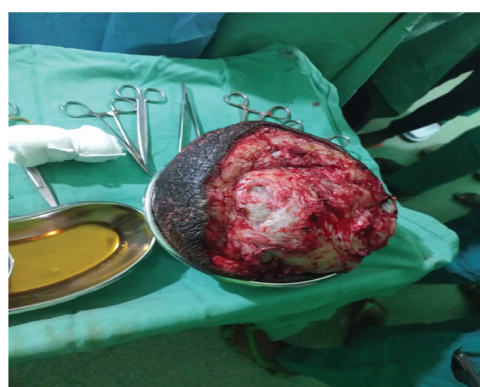


Fig. 3. The excised scrotal tissue.

therapies). He was placed on a 4-monthly cyclical administration of Ivermectin 12 mg stat, Albendazole 400 mg daily for 6 weeks, Doxycycline 200 mg stat, then 100 mg 12 hourly for 6 weeks. Surgically, he had a reduction scrotoplasty which involved intra-operative early identification of the testes with subsequent freeing and isolation of the testes from the gubernaculum (see Fig. 2); development of scrotal flaps on both sides of the scrotum; excision of the redundant scrotal tissue; and apposition of the scrotal flaps.

The scrotal skin and subcutaneous tissue were found to be thickened, measuring about 2 cm in thickness. Both testes and spermatic cords looked healthy. Haemostasis was achieved with electrocautery and suture ligation. The excised scrotal tissue (skin and subcutaneous tissue) weighed 4.4 kg (see Fig. 3). The postoperative period was uneventful and non-absorbable skin sutures were removed after 2 weeks. He was seen during his first follow-up visit and found to be very lively and happy with normal mobility and a good surgical scar (Figs. 4a and 4b). Histology of the excised tissue revealed extensive fibrosis with lymphostasis. He was to do a repeat seminal fluid analysis during the next visit but was unfortunately lost to follow-up.

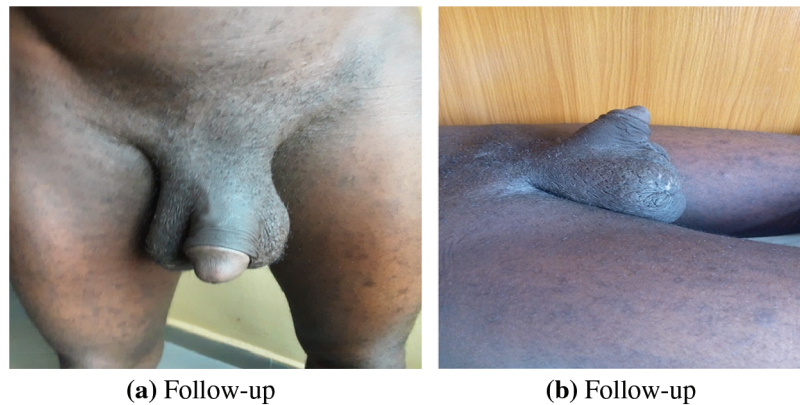


Fig. 4. (a) Follow-up. (b) Follow-up.

### 3. DISCUSSION

Lymphatic filariasis results from the obstruction to the free flow of lymphatic fluid through the lymphatic channels, a consequence of chronic inflammatory response to the dying worms in the afferent lymphatics and lymph node sinusoids draining the affected area. Isolated scrotal oedema is a rare clinical presentation of lymphatic filariasis even in endemic regions [3], [6]. The index patient was in his 4<sup>th</sup> decade of life which fell within the documented presenting age range of 26–64 years in other studies [3], [6], [7]. His age at presentation agrees with the belief that lymphatic filarial infections occur in childhood but present in adulthood following inflammatory response to dead and dying adult filarial worms. Our patient's clinical presentation was suggestive of the diagnosis which was confirmed by positive peripheral blood film for microfilaria of *W. bancrofti*. This pattern of suggestive clinical presentation with positive appropriately-timed peripheral blood films (12 noon, 10 pm, and 2 am) has been reported in endemic regions of Brazil but differs from the findings of other researchers in India, Ghana, and Indonesia where positive microfilaria peripheral blood film could not be demonstrated in similar patients [3], [6]–[8]. Thus, the diagnosis of filarial infection in these patients was based on suggestive clinical history, physical findings, and other laboratory investigations other than positive peripheral blood film for microfilaria. This failure to demonstrate microfilaria in the peripheral blood film could be due to prior administration of microfilaricidal drugs and, possibly, non-consideration of blood periodicity of microfilaria during blood sampling for microfilaria parasite [8].

The index patient presented after 10 years of enduring the psychosocial impact of the disease probably due to Ignorance, poverty, fear of stigmatization, and lack of specialists to make prompt and precise diagnoses of the disease. This delayed presentation seen in our patient is in tandem with the 2–15 years duration of illness at presentation reported in other studies and re-echoes why the disease is still listed among the neglected tropical diseases [3], [6]. Delay in presentation has been demonstrated to be frequently associated with huge psychosocial negative impact and recurrent bacterial infections of the lymphoedematous skin as reported in our patient and other studies [3], [6]. The disease is more prevalent among individuals of low socioeconomic class [3], [6], [9]. This was the case with

the index patient who was a peasant farmer who dropped out of tertiary education on account of the psychosocial impact of the disease. On account of the huge scrotal mass, he had difficulty ambulating, sexual dysfunction, and low self-esteem which impacted so much on his social well-being similar to what was observed in other studies [3], [6], [7]. Azoospermia was noted in the index patient as a complication of the chronic lymphoedematous state of the scrotum similar to the report by Andrade-Rocha and Cardona Maya [10] on a 21-year old man in Brazil with scrotal lymphoedema of 10 years. These must have arisen from the alteration in the thermoregulatory function of the scrotum and/or the pressure effect of the giant scrotum on the testes.

Treatment of filarial lymphedema of the scrotum involves non-surgical and surgical modalities. The non-surgical treatments are the use of pharmacologic agents and conservative therapy which includes skin hygiene, manual lymph drainage, compression bandages, and therapeutic exercises [11], [12]. Conservative therapy is often associated with poor outcomes and recurrence. Surgery remains the gold standard of treatment, especially in severe cases, and involves either lymphangioplasty or lymphangiectomy with reconstruction [11]–[14]. Several surgical techniques have been described, including staged surgeries, all geared towards achieving a good outcome [4], [14]. The excised scrotal tissue in our patient weighed 4.4 kg which is much lower than the 32 and 36 kg reported by Thejeswi *et al.* [3] in India and Tillyashaykhov *et al.* [15] in Uzbekistan, respectively. Our patient's mobility, self-esteem, and social well-being markedly improved following the surgery similar to the findings from other studies [4], [15]. Unfortunately, we could not follow up on his azoospermia as he absconded from further clinic visits probably due to financial constraints. Vives *et al.* [4] also documented a similar loss of their patient to follow-up due to lack of finance.

### 4. CONCLUSION

In conclusion, isolated scrotal lymphoedema from filariasis, though rare, could be debilitating with far-reaching organic, psychological, and economic implications. Good treatment outcomes hinge on appropriate medical and surgical interventions.

## CONFLICT OF INTEREST

Authors declare that they do not have any conflict of interest.

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