Filariasis with Squamous Cell Carcinoma: A Hidden Surprise

Abstract

Squamous cell carcinoma (SCC) arising with chronic lymphedema is a rare condition, though literature suggests a variety of malignant tumors associated with filariasis. We present a case of 70-year-old male patient with a history of penile and scrotal SCC of filarial origin. We here discuss the unusual association of carcinoma and filariasis and its surgical treatment.

Keywords: Chronic lymphedema, filariasis, squamous cell carcinoma

Introduction

Lymphatic filariasis, a neglected tropical diseases and ranked the second leading cause of disability in the world by the World Health Organization,[1] is caused by thread-like parasites mainly by Wuchereria bancrofti and Brugia malayi transmitted by Culex, Anopheles, Aedes, or Mansonia mosquitoes. It mainly affects the lymph nodes and lymphatic channels of lower limbs, spermatic cords, epididymis, and retroperitoneum and presents as swelling of the involved region when the adult worm blocks the lymphatic vessels. Cellular infiltration together with hyperplasia of the lymphatic endothelium occurs with repeated inflammatory episodes which results in painful and disfiguring manifestation such as lymphedema (elephantiasis), penile, and scrotal swelling. Extranodal sites such as the breast are rarely involved where they present as lump and sometimes mimic breast carcinoma.^[2] Chronic lymphedema is found to be associated with malignant changes which include Kaposi's sarcoma, melanoma, lymphoma, malignant fibrous histiocytoma, and squamous cell carcinoma (SCC).[3] In our case, chronic history of elephantiasis for about three decades leads to the development of SCC. SCC is a life-threatening entity and early diagnosis can lead to its control or cure.

Case Report

A 70-year-old male resident of Wardha district of Maharashtra presented with swelling of the scrotum and penis with a recurrent history of fever and lymphangitis.

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For 30 years, he also developed edema of the right lower limb. The patient was a known case of filariasis, and with the course of time, he suffered recurrent episodes of inflammation due to which there was progressive increase in the size of the scrotum and penis. The patient neglected the symptoms until it started interfering with his normal routine work following which he presented to the surgery department.

On general examination, the patient was obese and hypertensive but nondiabetic; other findings were normal. On systemic examination, cardiovascular system and respiratory systems were normal. The abdomen was soft and nontender on palpation, and there was no organomegaly.

Local examinations revealed large thickened, firm filarial scrotum having nonpitting skin along with Ram's horn penis [Figure 1] which had thickened glans, and the prepuce was nonretractile. The right lower limb had non-pitting lymphedema and minimal verrucae at the toes [Figure 2]. The presence of fungal infection was noted at the site. The inguinal lymph nodes were 2–3 in number, firm, and nontender on palpation.

Hematological and biochemical investigations were normal. Examination of scrotal aspirate revealed microfilariae [Figures 3 and 4]. Under anesthesia, scrotectomy was performed. Preputial skin was opened dorsally, and to our surprise, a cauliflower-like growth was seen over the glans involving the corona and part of the prepuce of 2.5 cm × 2.5 cm infiltrating on to the shaft of the penis [Figure 5]. Fine-needle aspiration cytology taken from

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both sides of the inguinal lymph nodes was negative for any malignancy. The frozen section confirmation of growth biopsy revealed it to be a case of SCC.



Figure 1: Inspection of genitals showing Ram's horn penis and scrotal swelling

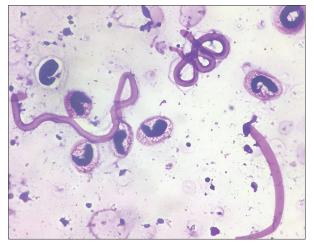


Figure 3: Scrotal aspirate showing microfilariae (low power). Courtesy: Vishal Kulkarni

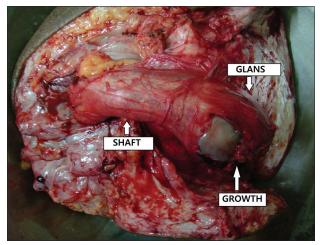


Figure 5: Growth over the enlarged glans and preputial skin

After proper explanation to the patient's relatives and with due consent, total amputation of the penis was performed [Figure 6], and the perineal urethral opening was created. The wound was closed keeping drain and catheter



Figure 2: Edema of the right lower limb



Figure 4: Scrotal aspirate showing microfilaria (high power). Courtesy: Vishal Kulkarni

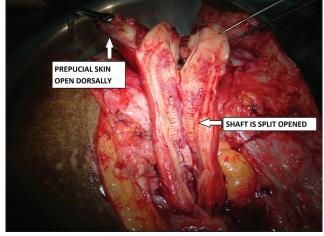


Figure 6: Amputation of penis and scrotectomy

in situ. The patient made an uneventful postoperative recovery, being discharged from the hospital after the surgery.

Two weeks postoperation, the patient was re-evaluated for any inguinal metastasis, which came out to be negative after which he was put on regular follow-up.

Discussion

Malignancy, mainly SCC, arising in chronic lymphedema is a rare entity having only nine cases reported, [4] majority of which are related to the limbs and breasts. [2,4,5]

Most of the reports in the literature suggest carcinogenesis in lymphedema to be associated with immunologic factors. Chronic lymphedema causes extrusion of protein into interstitial space with the influx of inflammatory mediators. Chronic inflammation is hypothesized as one of the major contributing factors for the development of malignancy due to poor hygienic conditions with infection following the involvement of bacteria into the edematous tissue. [6] The early recognition of tumor-specific antigen is hindered by a shortfall in afferent lymphatic drainage. [7]

This case emphasizes the need for mindful evaluation and follow-up of patients with chronic lymphedema. The possibility of a malignant growth in the lymphedematous tissue should be kept in mind. Chronic warty hyperkeratosis and fissuring may hide a lesion leading to delay in the presentation of a malignant ulcer. A dermatological opinion should be taken as and when necessary. Serum tumor markers for penile SCC such as SCC antigen are considered as link between these two rare conditions and could play a crucial role in the detection of such complex cases.^[8,9]

Conclusion

Patients should be instructed on good hygiene and regular monitoring of any affected area. Any new or suspicious growth should be examined and biopsied without delay.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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