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Case Report

A Rare Case of Large Unilateral Vulval Elephantiasis Involving Right Labia Majora

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Abstract

Genital elephantiasis of the female is very rare. Filariasis is the most common cause of lymphedema of the female genitalia; other causes are lymphogranuloma venereum, granuloma inguinale, carcinoma, and tuberculosis. Mrs. X, 25-year-old female, came with complaints of huge progressively increasing swelling involving the right labia majora for 4 years and associated with difficulty in walking. Past history of fever for 6 months and treated with diethylcarbamazine (DEC). On examination, non-ulcerative, polypoidal growth of 30 × 25 × 15 cm involving the right labia majora, obstructing the vulval cleft. No associated lymphadenopathy or limb edema. All investigations were within normal limits. The patient was given DEC and excision and primary closure were done. Biopsy showed skin with intraepidermal and subepidermal clearing enclosing eosinophilic fluid with scattered lymphocytes and subepithelium showing dense fibrosis and inflammatory infiltrates suggestive of chronic inflammatory pathology. In endemic countries like India, Early diagnosis and appropriate treatment prevent disfigurement and its sequelae.

Key words: Filariasis, Vulval elephantiasis, Vulval lymphedema

INTRODUCTION

Vulval elephantiasis is the lymphedema of soft tissues that develops following the accumulation of protein-rich interstitial fluid secondary to improper lymphatic drainage of the involved area. In endemic countries, filariasis is the most common cause of lymphedema, which commonly affects the lower limbs, arms, and rarely involve genital organs. Filarial elephantiasis of the vulva in females accounts for only 1–2%.[1]

CASE DESCRIPTION AND RESULT

A 25-year-old female, P1L1, came to our department with complaints of large pendulous swelling in the vulval region for 4 years, associated with difficulty in walking, difficult to maintain genital hygiene and sexual life [Figure 1]. She had a past history of fever for 6 months back and treated with diethylcarbamazine (DEC). Local examination showed a large pendulous, mass of hypertrophied tissue of 30 × 25 × 15 cm hanging down and obstructing the vulval cleft on the right labia majora. Overlying skin was hard and thickened and showed extensive rugosities [Figure 2]. The overlying skin was chronically infected with areas of ulceration and pigmentation. On palpation, there was no warmth and tenderness; swelling was firm and non-pitting. No inguinal lymphadenopathy or any scars and sinuses in that area. All the investigations were normal. Magnetic resonance imaging showed hyperintense subcutaneous edema noted in the right labia majora with few internal echoes showing no diffusion restriction, associated diffuse skin thickening of maximum thickness of 1.25 cm, focal areas of skin noted in the anteroinferior aspect possibility of ulceration and above mentioned features suggesting possibility of elephantiasis of right labia majora and no evidence of intrapelvic extension. No significant inguinal lymph node [Figure 3]. DEC in the dose of 6 mg/kg/day in three divided doses was given for 21 days. Under spinal anesthesia, lithotomy position, parts painted and draped. Markings were made around the swelling, an incision was made, and the incision was deepened and dissection was done. Swelling excised preserving features of the labia majora [Figure 4].

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Perfect hemostasis was achieved and the remaining skin edges were sutured using 3.0 cutting prolene and a sterile dressing done [Figure 5]. Uneventful post-operative period. Histopathological report showed skin with intraepidermal and subepidermal clearing enclosing eosinophilia fluid with scattered lymphocytes and subepithelium showing dense fibrosis and inflammatory infiltrates composed of lymphocytes, plasma cells, and congested blood vessels, suggestive of a chronic inflammatory condition probably filariasis. Post-operatively DEC in the dose of 6 mg/kg in three divided doses was given for 21 days. Patient was discharged on post-operative day 5. Patient was able to walk without any difficulty, able to maintain genital hygiene and sexual life.

Figure 1: Vulval elephantiasis involving the right labia majora

Figure 2: Preoperative image showing incision marking

DISCUSSION

Genital elephantiasis due to filariasis is very rare, and it accounts for only 1–2% of filariasis.[1,2] Other causes of genital lymphedema include Lymphogranuloma venereum, granuloma inguinale, carcinoma, tuberculosis, and chronic infections.[3,4] Lymphedema occurs due to an inability of the existing lymphatic system to accommodate the protein and fluid entering the interstitial compartment at the tissue level. Lymphedema is classified into primary and secondary lymphedema. The cause is unknown in primary lymphedema, whereas secondary lymphedema is due to known causes, such as infections and malignancies. The primary lymphedema with onset before 2 years of age is referred to as congenital, the familial version of which is known as Milroy's disease. Primary lymphedema with onset between 2 and 35 years of age is called lymphedema praecox, which is the most common form of primary lymphedema. The familial version of lymphedema praecox is known as Meige's disease.[5] Primary lymphedema with onset after 35 years of age is called lymphedema tarda. Lymphedema occurs in three stages. In the first stage, impaired lymphatic result in protein-rich accumulation in the interstitial compartment and manifest as soft-pitting edema. The second stage includes accumulation of fibroblasts and macrophages in the affected tissues, resulting in a local inflammatory response and deposition of connective tissue in the skin and subcutaneous layer, and formation of non-pitting edema. In the third stage, repeated infection and local inflammatory response

Figure 3: Magnetic resonance imaging showing subcutaneous edema and diffuse skin thickening

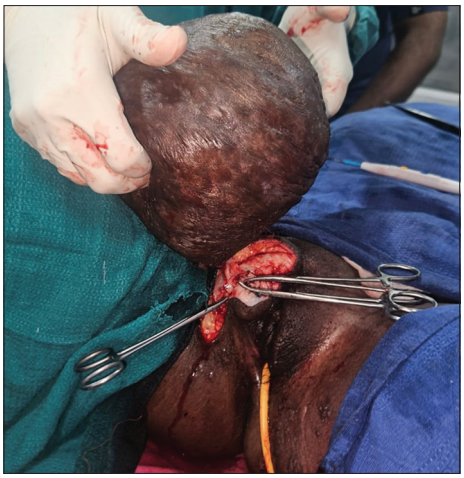

Figure 4: Excision of swelling

Figure 5: Final suture line

result in excessive subcutaneous fibrosis and scarring with severe skin changes characteristic of lymphatic elephantiasis.[5] The most common form of lymphedema is secondary lymphedema. In developed countries, the most common causes of secondary lymphedema involve resection or ablation of the regional lymph nodes by surgery, radiation, tumor invasion, direct trauma, or, less commonly, an infectious process. Globally, filariasis, caused by the infestation of the lymph nodes by the parasite Wuchereria bancrofti, is the most common cause of secondary lymphedema. Infection is usually acquired early in childhood, but it may take several years to manifest. The most severe symptoms of chronic disease are generally more common in males than in females. In Bancroftian filariasis, the main clinical features are hydrocele, lymphedema, elephantiasis, and chyluria, whereas for Brugian filariasis, the urogenital areas are commonly spared. Investigations, such as detection of microfilaria in night blood specimens, detection of circulating filarial antigens (CFA), and detection of filarial DNA in human blood by polymerase chain reaction. Antigen testing is most widely used for diagnosis. CFA detection test is the "gold standard" for diagnosing Wuchereria bancrofti infections. Elephantiasis is a preventable morbidity. The Global Program to Eliminate Lymphatic Filariasis coordinated by the World Health Organization, aims to eliminate lymphatic filariasis (LF) by 2020 using repeated rounds of mass drug administration. In the filariasis control program (1955) which has now been integrated as the "elimination of LF program" under the National Vector Borne Diseases Control Program, mass prophylaxis for filariasis is given annually in the form of DEC. Annually, a single dose of one drug (ivermectin or DEC) administered can result in a 90% reduction and single-dose co-administration of two drugs, Ivermectin+DEC or albendazole, reduces blood microfilaria by 99% for a full year.[1] Hydrocele and genital elephantiasis have to be managed surgically.

CONCLUSION

Vulval elephantiasis is very rare manifestation of filariasis, which is endemic in our country. Mass prophylaxis under the filariasis control program can reduce its endemicity and sequelae. Early detection and treatment prevent the complications and disfigurement caused by filarial infection. Management of vulval elephantiasis includes a multidisciplinary approach involving Gynecologist, Dermatologist, and Plastic surgeon. Surgical treatment is necessary for the treatment of Genital elephantiasis, which causes disfigurement.

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