Acquired Cutaneous Lymphangiectasia of the Scrotum Secondary to Filarial Lymphoedema

M P Binithaa, Anza Khadera, Pentam Beegum Sherjeenaa, M R Rinia

a. Department of Dermatology and Venereology, Government Medical College, Kozhikode*

ABSTRACT

Published online on 30th December 2015

A middle-aged man developed acquired cutaneous lymphangiectasis of the scrotum secondary to filarial lymphedema. He had distressing copious watery discharge, which was completely relieved by carbon dioxide laser therapy

Keywords: Acquired cutaneous lymphangiectasia, Scrotum, Filariasis

INTRODUCTION

Lymphangiectasia or acquired lymphangioma is characterised by the formation of cutaneous vesicles containing lymph as a result of the obstruction of the lymphatic vessels due to various causes. Though filariasis is a common cause of lymphatic obstruction and lymphedema in our region, reports of acquired

Figure 1. Lymphedema of the scrotum and left lower limb

cutaneous lymphangiectasia (ACL) occurring on the scrotum are very rare. We present such a case.

CASE REPORT

A 52-year-old man presented with asymptomatic vesicles on the scrotum exuding clear fluid, of six months duration. He was in great discomfort due to the constant dribbling and wetting of his clothing. Following an episode of filariasis two years ago, he had developed persistent swelling of the left leg which later progressed to involve the whole of the left lower limb. He gave a history of recurrent episodes of fever, pain and inflammatory swelling of the left leg suggestive of cellulitis.

Systemic examination was within normal limits. Dermatological examination revealed diffuse non-pitting oedema of the whole of the left lower limb (figure 1).



Figure 2. Fluid-filled vesicles on the scrotum.

Corresponding Author:

Dr. M P Binitha, Professor, Department of Dermatology and Venereology, Government Medical College, Kozhikode. Email: mpbinitha56@gmail.com

^{*}See End Note for complete author details



Figure 3. Vesicles on the scrotum oozing clear fluid

The skin of the scrotum was thickened, oedematous and diffusely covered with small vesicles containing clear fluid (figure 2). There was profuse oozing from the vesicles (figure 3).

Routine investigations including blood cell count, erythrocyte sedimentation rate, biochemical values, urine analysis, eosinophil count and serum IgE level were within normal limits. Microscopic examination of a blood smear was negative for microfilariae.

A biopsy taken from a vesicle on the scrotal skin revealed mild hyperkeratosis and acanthosis. The papillary and upper dermis contained multiple dilated lymph vessels lined by a single layer of flattened endothelial cells, containing clear fluid and occasional red blood cells in the lumina. There were no smooth muscle cells around the cisternae. The deep dermis and subcutaneous tissue were unremarkable except for a mild inflammatory infiltrate.

The patient was treated with pulsed carbon dioxide (CO2) laser tissue ablation at weekly intervals. After six sittings, there was complete subsidence of the lesions.

DISCUSSION

Lymphangiomas are lymphatic malformations which may be congenital or acquired. Congenital lymphangiomas or lymphangioma circumscriptum (LC) appear on the skin as localised swellings, with varying degrees of deeper involvement. Acquired lymphangiomas (AL) are also called lympangiectases and occur as a result of obstruction to lymphatic vessels in the deeper layers of the skin, causing retrograde pressure and dilatation of the lymphatics in the upper dermis.

Congenital lymphangiomas are not a component of the normal lymph channel system. Lymphangi-

oma circumscriptum is a vascular malformation which is a true localised hamartoma of the skin. It usually presents at birth, in infancy or childhood, though adults may rarely be affected. The lymphatics in the deep dermis and subcutaneous tissue are dilated in LC, frequently forming large lymphatic cisterns and extensive deeper involvement. A combined lymphaticovenous malformation or bleeding into the cysts have also been observed. Secondary changes of the surface may be seen, giving rise to a warty appearance. Histopathologically, the lymph cisterns in LC may have smooth muscle cells in their walls, a significant finding which is absent in AL. Treatment is often difficult due to extensive subcutaneous cystic channels, and total surgical excision may not be feasible.

Lymphangiectasis or AL are seen in adults, usually in the fifth or sixth decade of life. They may be localised or extensive, and situated on the limbs, vulva and other areas. Lymphangiectasis present as grouped or dispersed vesicles usually on a lymphedematous area, although rarely the underlying skin may appear normal. Sometimes the vesicular nature may not be apparent and they may present as small, smooth nodules. A history of an aetiological event is usually present in AL. The lymph stasis occurs as a result of secondary processes which cause blockage of the originally normal lymph drainage, such as scarring and destruction of the deeper lymphatic vessels or obstruction of the draining lymph nodes. Damage to the lymphatics most often follows surgery for malignancy which involves extensive dissection of the deeper tissues such as radical mastectomy,1 as well as radiotherapy alone or following surgery for cancer,2 regional lymph node obstruction due to mass lesions, tuberculosis, filariasis, lymphogranuloma venereum, metastases, and scarring processes.4 Histopathologically AL involves only the superficial lymphatic vessels. Though a few red blood cells may be present in the lymphatics, frank bleeding is usually absent.

Filariasis is a common cause of lymphedema in the tropical countries. Lymphedema of the lower limbs often involves the scrotum as well. However, filariasis leading to lymphangiectasis has been very rarely reported. Overall, very few reports of lymphangiectasis of the scrotum and penis are available, and are due to other causes such as primary lymphedema, and following vasectomy.

Complications include secondary bacterial infection and recurrent cellulitis, as well as cosmetic disfigurement and emotional distress due to the watery discharge. Rarely, squamous cell carcinoma¹⁰ and

lymphangiosarcoma¹¹ (Stewart-Treves syndrome) may occur. Development of keloids following CO2 laser treatment for vulval LC has also been reported.²

Treatment includes local care to prevent secondary bacterial infection of the ruptured vesicles by cleansing and application of antibiotic ointments. Surgical modalities include electrodessication, sclerotherapy, cryotherapy and surgical excision. Vulvar lesions have been demonstrated to show a good response to CO2 laser therapy. Our case demonstrates that lymphangiectasis on the scrotum are also highly amenable to this form of treatment, with a dramatic improvement in the quality of life of the patient.

END NOTE

Author Information

- Dr. M P Binitha, Professor,
 Department of Dermatology and Venereology,
 Government Medical College, Kozhikode.
- Dr. Anza Khader, Assistant Professor,
 Department of Dermatology and Venereology,
 Government Medical College, Kozhikode
- Dr. Pentam Beegum Sherjeena, Senior Resident, Department of Dermatology and Venereology, Government Medical College, Kozhikode
- Dr. M R Rini, Junior Resident, Department of Dermatology and Venereology, Government Medical College, Kozhikode

Conflict of Interest: None declared

Cite this article as: Binitha MP, Khader A, Sherjeena PB, Rini MR. Acquired Cutaneous Lymphangiectasia of the Scrotum Secondary to Filarial Lymphoedema. Kerala Medical Journal. 2015 Nov 27;8(4):29–31

REFERENCES

- Tasdelen I, Gokgoz S, Paksoy E, Yerci O, Cetintas SK, Demiray M, et al. Acquired lymphangiectasis after breast conservation treatment for breast cancer: report of a case. Dermatol Online J. 2004;10(1):9.
- Jappe U, Zimmermann T, Kahle B, Petzoldt D. Lymphangioma circumscriptum of the vulva following surgical and radiological therapy of cervical cancer. Sex Transm Dis. 2002 Sep;29(9):533–5.
- Di Leonardo M, Jacoby RA. Acquired cutaneous lymphangiectasias secondary to scarring from scrofuloderma. J Am Acad Dermatol. 1986 Apr;14(4):688–90.
- Russell B. Lymphangioma circumscriptum and keloids. Br J Dermatol. 1951 Apr;63(4):158–9.
- Hagiwara K, Toyama K, Miyazato H, Nonaka S. A case of acquired lymphangioma due to a suspected old filariasis and a review of literature. J Dermatol. 1994 May;21(5):358–62.
- Weakley DR, Juhlin EA. Lymphangiectases and lymphangiomata. Arch Dermatol. 1961 Oct;84:574

 –8.
- Haroon S, Hasan SH. Lymphangioma circumscriptum in the scrotum: a case report. J Med Case Rep. 2012;6:233.
- Suehiro K, Morikage N, Murakami M, Yamashita O, Hamano K. Primary lymphedema complicated by weeping chylous vesicles in the leg and scrotum: report of a case. Surg Today. 2012 Nov;42(11):1100–3.
- Pal DK, Banerjee M, Moulik D, Biswas BK, Choudhury MK. Lymphangioma circumscriptum of the scrotum following vasectomy. Indian J Urol. 2010 Apr;26(2):294–5.
- Short S, Peacock C. A newly described possible complication of lymphangioma circumscriptum. Clin Oncol (R Coll Radiol). 1995;7(2):136–7.
- King DT, Duffy DM, Hirose FM, Gurevitch AW. Lymphangiosarcoma arising from lymphangioma circumscriptum. Arch Dermatol. 1979 Aug;115(8):969–72.