Received: 2 December 2021 | Revised: 19 March 2022 | Accepted: 13 April 2022

DOI: 10.1002/ccr3.5839

CASE REPORT

Clinical Case Reports Open Access WILEY

Cutaneous lymphangioma secondary to breast cancer radiotherapy after mastectomy: A case report

Safoura Shakoei[1] | **Maryam Noorbakhsh Sabet**[2] | **Alireza Ghanadan**[3]

[1]Department of Dermatology, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran

[2]Department of Dermatology, Razi Hospital, Tehran University of Medical Sciences (TUMS), Tehran, Iran

[3]Department of Pathology, Imam Khomeini Hospital, Tehran University of Medical Sciences, Tehran, Iran

Correspondence
Safoura Shakoei, Department of Dermatology, Imam Khomeini Hospital, Tehran University of Medical Sciences (TUMS), Tehran PC:1419733141, Iran.
Email: dr.shakoei@gmail.com

Funding information
None

Abstract

A 60-year-old woman presented with non-itchy shiny translucent papules on her right breast surface. She had a history of quadrantectomy and chemoradiotherapy due to medullary breast carcinoma 20 years ago. Cutaneous lymphangioma occurred secondary to surgery and radiotherapy 8 years ago. It is a late complication of breast cancer treatment.

KEYWORDS

breast cancer, cutaneous lymphangioma, mastectomy, radiotherapy

1 | INTRODUCTION

Cutaneous lymphangioma (CL) is a rare benign disorder of lymphatic vessels that usually occurs in regions such as the head, neck, and oral cavity. Cutaneous lymphangioma primarily occurs in children, especially before the age of two.[1] From a clinical point of view, CL generally manifests as clusters of translucent papules and vesicles that are heterogeneous in color, ranging from colorless to purple. However, these malformations might occur secondary to some conditions such as radiation therapy. The data on the association between breast cancer radiation therapy and breast lymphangioma remains significantly limited.[2,3]

Due to its rarity, we describe a woman with a history of breast cancer and chemoradiotherapy with superficial lymphangioma on her right breast. It is a rare disease but could be easily diagnosed with histological examination.

2 | CASE PRESENTATION

A 60-year-old woman presented to our clinic with non-itchy shiny translucent papules on her right breast surface since 8 years ago. She had a history of medullary breast carcinoma 20 years ago, underwent quadrantectomy and axillary dissection, and had a history of chemotherapy and breast and supraclavicular regions radiotherapy in two tangential fields (lateral and medial) at a dose of 5000 centigray (cGy) in 25 fractions for the whole breast and a direct anterior-posterior field for the supraclavicular region at a dose of 5000 cGy in 20 fractions with Cobalt-60 Machine 20 years ago. She underwent a boost dose of 1000 cGy tumor bed in 5 sessions with the Siemens machine. The patient's lesions had not increased ever since.

A physical examination showed multiple raised, translucent papulovesicular lesions varying in size from a

pinhead to one cm over the chronic radiodermatitis area of the patient's right breast (Figure 1). No other symptoms or signs were found in the patient.

The patient underwent breast skin lesion biopsy with differential diagnoses of lymphangioma and metastasis. Histopathologic examination showed superficial widely-dilated lymphatic spaces lined with thin endothelial cells with uniform hyperchromatic oval to flattened nuclei. Lymphatic vessels contained some proteinaceous material and scattered lymphocytes around the vessels with hyalinized septae. There was also epidermal atrophy over the lymphatic channels and elongation and thinning of the rete ridges embracing lymphatic channels. A diagnosis of superficial lymphangioma was confirmed (Figure 2). The lesions were benign, and the patient refused to receive treatment. After explaining the purpose of the report to the patient, written informed consent was obtained from her.

3 | DISCUSSION

Cutaneous lymphangioma is an acquired vesicular dilation of lymphatic channels. Although it is a primary disease, it may occur secondary to some conditions. It has been shown that the development of CL is associated with the treatment of some cancers, such as breast and cervix cancer.[3,4] The list of treatment protocols that cause this condition includes surgery alone, irradiation alone, and combined surgery and irradiation.[5] Cutaneous metastasis (CM) of breast carcinoma rarely manifests as telangiectatic carcinoma. Lymphangioma circumscriptum, similar to telangiectatic carcinoma, is a rare form of CM and should be differentiated from it.[6]

Radiation therapy is considered a major strategy in the treatment of various types of cancer. However, irradiation can also cause some negative changes in tissues and lead to tissue complications. For instance, atypical vascular lesions (AVLs) are benign vascular proliferations that usually develop after radiation therapy. AVLs present as small papules or nodules in both solitary and multiple contexts.[7] These lesions generally exhibit two histologic patterns, including superficial lymphangioma-like and hobnail hemangioma-like/benign lymphangio-endothelioma-like patterns.[8] The biopsy of our case was consistent with the superficial lymphangioma-like pattern, which is caused by fibrosis formation in the junction of the fatty tissue and reticular dermis.

Radiotherapy induced fibrosis and lymphatic obstruction increase local pressure and consequently lead to lymphatic fluid accumulation.[2] Clinically, due to saccular dilatation of superficial lymphatic channels, lesions may vary from hyaline fluid vesicles to reddish nodules.[3] These lesions are mainly benign and different in size, depending on the extent of fluid collection. However, radiation therapy can provoke the development of some malignant lesions, such as angiosarcoma.[2]

Lymphocyte infiltration may be due to chronic irritation caused by intertrigo and maceration. The treatment of lymphangioma is mostly elective and for cosmetic and functional reasons. However, some secondary complications are probable such as infection and subsequent cellulitis. Several treatment modalities have been used, including electrodesiccation, laser therapy, cryotherapy, sclerotherapy, and surgical excision.[9] In our case, although we explained all of the benefits of treatment modalities (electrodesiccation, laser therapy, cryotherapy), the patient refused to receive any kind of treatment.

In conclusion, breast lymphangioma might occur secondary to radiation therapy. Although this phenomenon is a rare and late complication of radiotherapy, it should always be considered in the diagnosis of patients with vesicular lesions in the field of radiation therapy in the differential diagnosis of metastasis. Further reports are required to highlight this issue in an attempt to help oncologists and dermatologists diagnose patients referred with vascular lesions and a positive history of radiotherapy. In patients with a history of breast cancer, the disease is always a concern for physicians. Recognizing this late complication can be effective in reducing this concern.

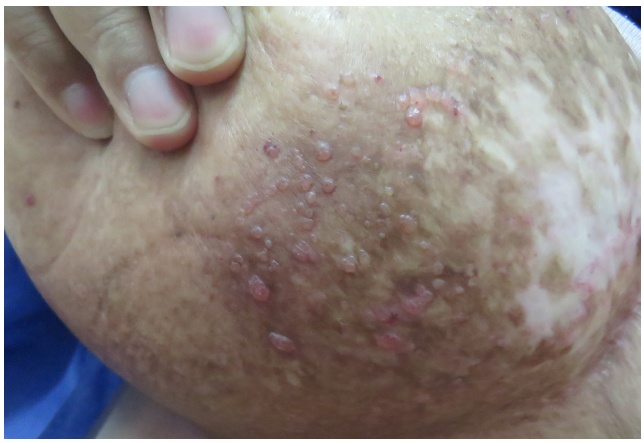

FIGURE 1 Multiple raised, translucent papulovesicular lesions over the chronic radiodermatitis area of the right breast

AUTHOR CONTRIBUTIONS

(1) Manuscript: A. Writing of first draft, B. Review, and Critique. (2) Pathology report. Safoura Shakoei, 1A, 1B. Maryam Noorbakhsh Sabet, 1A, 1B. Alireza Ghanadan, 1B, 2. All authors read and approved the final manuscript.

ACKNOWLEDGMENT

None.

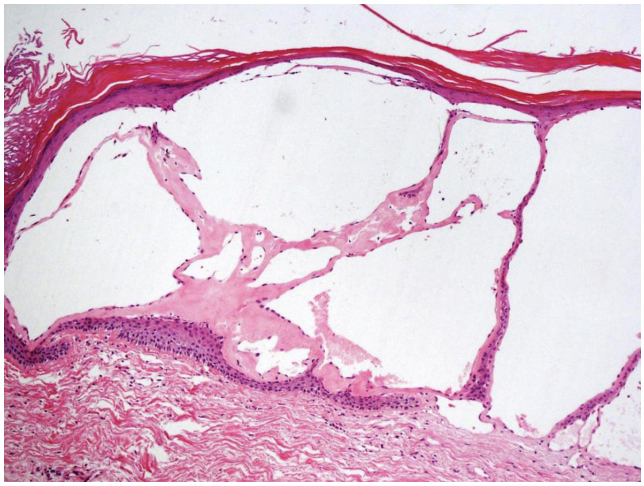

FIGURE 2 Cystically-dilated lymphatic vessels lined by thin endothelium with a uniform flat to oval nuclei separated by hyalinized fibrous septae in papillary dermis surrounded by elongated rete ridges and intersected by hyalinized septae (H&E × 10)

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICAL APPROVAL

We hereby confirm that the present study conforms to the ethical standards and guidelines of the journal.

CONSENT

The patient gave written informed consent for the online publication of her picture.

ORCID

Safoura Shakoei https://orcid.org/0000-0001-6790-7633

REFERENCES

1. Orvidas LJ, Kasperbauer JL. Pediatric lymphangiomas of the head and neck. *Ann Otol Rhinol Laryngol*. 2000;109(4):411-421.
2. Oliveti A, Biasi TB, Funchal GDG. Lymphangioma secondary to irradiation after mastectomy. *An Bras Dermatol*. 2017;92(3):395-397.
3. Kaya TI, Kokturk A, Polat A, Tursen U, Ikizoglu G. A case of cutaneous lymphangiectasis secondary to breast cancer treatment. *Int J Dermatol*. 2001;40(12):760-761.
4. Ramli I, Ismaili N, Amarouch H, Senouci K, Hassam B. Vulvar lymphangioma circumscriptum following pelvic radiotherapy. *Presse Med*. 2016;45(3):374-375.
5. Chiyomaru K, Nishigori C. Acquired lymphangiectasia associated with treatment for preceding malignant neoplasm: a retrospective series of 73 Japanese patients. *Arch Dermatol*. 2009;145(7):841-842.
6. Bayramgürler D, Demirsoy EO, Doğan N, Yıldız K, Utkan Z. Lymphangioma circumscriptum-like presentation of breast carcinoma. *Indian J Dermatol Venereol Leprol*. 2012;78(1):121.
7. Patton KT, Deyrup AT, Weiss SW. Atypical vascular lesions after surgery and radiation of the breast: a clinicopathologic study of 32 cases analyzing histologic heterogeneity and association with angiosarcoma. *Am J Surg Pathol*. 2008;32(6):943-950.
8. Weaver J, Billings SD, editors. Postradiation cutaneous vascular tumors of the breast: a review. *Semin Diagn Pathol*. 2009;26(3):141-149. Elsevier.
9. Tasdelen I, Gokgoz S, Paksoy E, et al. Acquired lymphangiectasis after breast conservation treatment for breast cancer: report of a case. *Dermatol Online J*. 2004;10(1):9.

How to cite this article: Shakoei S, Noorbakhsh Sabet M, Ghanadan A. Cutaneous lymphangioma secondary to breast cancer radiotherapy after mastectomy: A case report. *Clin Case Rep*. 2022;10:e05839. doi:10.1002/ccr3.5839