

Non-invasive diagnosis of acquired lymphangiectases using optical coherence tomography

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Non-invasive diagnosis of acquired lymphangiectases using optical coherence tomography

Dear Editor,

Acquired lymphangiectases are permanent dilatations of cutaneous lymphatic vessels, which result from damage to and/or obstruction of previously normal lymphatic vessels by external causes such as surgery, trauma, radiotherapy and/or infections.¹⁻⁶ Clinically, acquired lymphangiectases appear as clusters of asymptomatic, translucent, flesh-coloured vesicular lesions; some of which may turn purple due to the presence of erythrocytes.^{1,7} Dermoscopy features include well-circumscribed, white-yellowish lacunae surrounded by pale septa with sometimes scattered reddish areas and red lacunae.⁷ Acquired lymphangiectases can mimic various dermatological disorders, namely angiokeratoma, basal cell carcinoma (BCC), cutaneous metastases, warts and immunobullous diseases, so histopathological examination is sometimes needed.^{8,9}

Optical coherence tomography (OCT) is a non-invasive diagnostic method with a penetration depth around 1.0-1.5mm, producing

real-time, in vivo, cross-sectional images of skin lesions.¹⁰ OCT is used for various indications, such as diagnosis of BCC, psoriasis, immunobullous diseases and haemangiomas.¹¹⁻¹⁴

We describe and correlate with histology the OCT features of two cases of acquired lymphangiectases occurring after radical mastectomy and trauma, respectively. A commercially available OCT system was used for Imaging (VivoSight, Michelson Diagnostics, Kent, UK, axial resolution < 5µm, lateral resolution < 7.5 µm).

Case 1 concerns a 50-year-old female patient who presented with a 5-mm erythematous shiny papule on the central abdomen (Figure 1A), which developed two months earlier. In 2011, she was treated for breast cancer with radical mastectomy without adjuvant radiotherapy and developed lymphedema in the left arm. The clinical differential diagnosis included nodular BCC and folliculitis. The OCT image showed well-defined roundish black (areflective) cavities separated by a septum, extending along the epidermis and

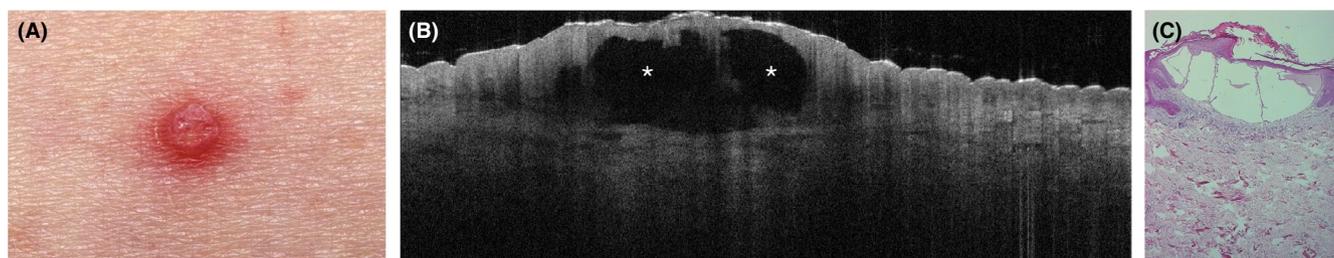


FIGURE 1 A, Clinical image of patient 1. B, OCT image of patient 1, revealing well-defined roundish black (areflective) cavities (asterisk) in the epidermis reaching to the papillary dermis, separated by a septum. C, Histopathology image of patient 1. Magnification 50x [Colour figure can be viewed at wileyonlinelibrary.com]

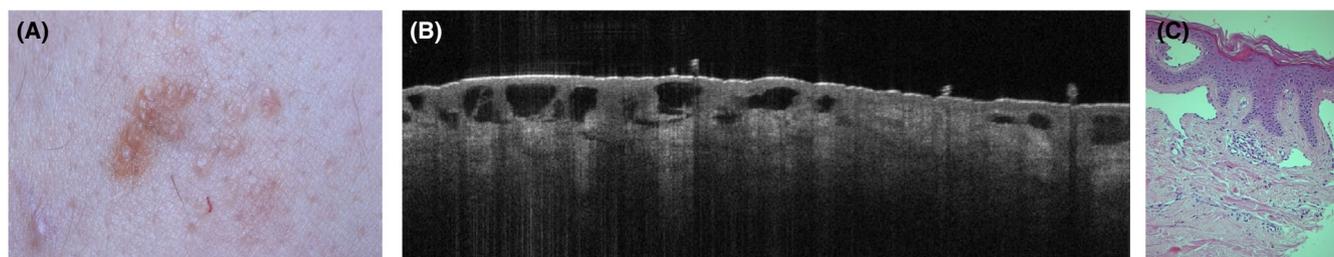


FIGURE 2 A, Clinical image of patient 2. B, OCT image of patient 2, showing well-defined roundish black (areflective) cavities of various size dispersed in the epidermis and papillary dermis. C, Histopathology image of patient 2. Magnification 200x [Colour figure can be viewed at wileyonlinelibrary.com]

reaching into the papillary dermis (Figure 1B). After punch biopsy, histology was assessed by a dermatopathologist and showed a cavernous epidermal lesion reaching to the papillary dermis, elevating the epidermis above the general level of the skin. It consists of grouped dilated lymphatic vessels that abut closely on the overlying epidermis and are thin walled with predominantly one layer of endothelial lining. The vessels contain proteinaceous lymph and few erythrocytes and inflammatory cells. No atypia was observed. (Figure 1C).

Case 2 concerns a 21-year-old male patient who presented with a lesion that developed 8 months after an open wound caused by a bicycle accident, and now showed a lenticular area of grouped yellow-brown vesicles with red lobules on the dorsal side of the right elbow (Figure 2A). The differential diagnosis consisted of acquired lymphangiectasis. On OCT, well-defined roundish black (areflective) cavities were seen of various size dispersed in the epidermis, reaching to the papillary dermis (Figure 2B). Histological assessment revealed dilated lymphatic vessels, dispersed in the lower epidermis and papillary dermis (Figure 2B). The vessels contain proteinaceous lymph, and a few erythrocytes are observed.

Based on the clinical and histopathological findings, the dermatopathologist established the diagnosis acquired lymphangiectases in both cases.

There is good correlation between OCT features and histopathology. With OCT, acquired lymphangiectases can be clearly visualized as well-defined roundish black (areflective) cavities in the epidermis reaching to the papillary dermis. These OCT images show resemblance to that of haemangiomas which also show well-defined roundish cavities in the upper dermis, as previously described by Salvini et al.¹⁴ However, in haemangiomas, cavities appear grey (hyporeflective), due to the presence of erythrocytes, whereas in lymphangiectasias cavities appear black (areflective), as lymph shows no internal scattering at all.^{14,15}

We demonstrated that OCT can guide the clinical differential diagnosis. In our first patient, a nodular BCC was the most likely clinical diagnosis, which was directly excluded with OCT. On OCT, nodular BCC has typical features including roundish dark grey structures in the dermis with a bright halo, sometimes surrounded by a dark border.¹⁶ The presence of well-defined roundish black cavities was a clue for lymph containing vessels. A rare cutaneous lymphangiosarcoma that may complicate chronic lymphedema is a poorly defined dermal tumour which infiltrates subcutaneous fat and other tissues and often has a multifocal distribution, with irregular shaped vessels.¹⁷ As acquired lymphangiectases present with well-defined grouped vessels which expand from the epidermis into the papillary dermis, we expect that differentiation between both diagnoses on OCT is possible.¹⁸ However, when in doubt, OCT may be used to select a representative area for a biopsy. Advantages of OCT are its non-invasive character, the immediate result and the fact that the entire lesion can be visualized. The depth and diameter of both lymphatic and blood vessels can be measured. A limitation of OCT is the resolution, which is not high enough to visualize single cells.¹⁹

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REFERENCES

1. Requena L., Sangueza O.P. Cutaneous vascular anomalies. Part I. Hamartomas, malformations, and dilation of preexisting vessels. *J Am Acad Dermatol.* 1997;37(4):523–549; quiz 549–552.
2. 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.
3. Zhang R.Z., Yang Y.H., Zhu W.Y.. Acquired lymphangiectasia of the glans following circumcision. *J Dtsch Dermatol Ges.* 2014;12(7):623–624.
4. Rao A.G.. Acquired lymphangiectasis following surgery and radiotherapy of breast cancer. *Indian J Dermatol.* 2015;60(1):106.
5. Giannelli V., Rockley P.F.. Acquired lymphangiectasis following mastectomy and radiation therapy—report of a case and review of the literature. *Cutis.* 1996;58(4):276–278.
6. Chang M.B., Newman C.C., Davis MDP, et al. Acquired lymphangiectasia (lymphangioma circumscriptum) of the vulva: clinicopathologic study of 11 patients from a single institution and 67 from the literature. *Int J Dermatol.* 2016;55(9):e482–e487.
7. Verzi A.E., Lacarrubba F., Tedeschi A., et al. Localized acquired lymphangiectasias after breast surgery: Enhanced non-invasive diagnosis using dermoscopy and reflectance confocal microscopy. *Skin Res Technol.* 2020;26(2):205–208.
8. Sharma R., Tomar S., Chandra M.. Acquired vulval lymphangiectases mimicking genital warts. *Indian J Dermatol Venereol Leprol.* 2002;68(3):166–167.
9. Brothers R.J., Crowe D.R.. Acquired widespread lymphangiectasia mimicking immunobullous disease: a case report. *SAGE Open Med Case Rep.* 2018;6:2050313X18802137.
10. Cheng H.M., Guitera P.. Systematic review of optical coherence tomography usage in the diagnosis and management of basal cell carcinoma. *Br J Dermatol.* 2015;173(6):1371–1380.
11. Ulrich M., Braunmuehl T., Kurzen H., et al. The sensitivity and specificity of optical coherence tomography for the assisted diagnosis of nonpigmented basal cell carcinoma: an observational study. *Br J Dermatol.* 2015;173(2):428–435.
12. Gambichler T., Moussa G., Sand M., Sand D., Altmeyer P., Hoffmann K.. Applications of optical coherence tomography in dermatology. *J Dermatol Sci.* 2005;40(2):85–94.
13. Welzel J., Bruhns M., Wolff H.H.. Optical coherence tomography in contact dermatitis and psoriasis. *Arch Dermatol Res.* 2003;295(2):50–55.

14. Salvini C., Massi D., Cappetti A., et al. Application of optical coherence tomography in non-invasive characterization of skin vascular lesions. *Skin Res Technol.* 2008;14(1):89-92.
15. VivoSight, Michelson Diagnostics, Kent, UK. <https://vivosight.com/clinician/training-support/>. Accessed June 01, 2020.
16. Hussain A.A., Themstrup L., Jemec G.B. Optical coherence tomography in the diagnosis of basal cell carcinoma. *Arch Dermatol Res.* 2015;307(1):1-10.
17. Weedon D. *Weedon's Skin Pathology*. 2002. Third edition. 919-922.
18. Sharma A., Schwartz R.A.. Stewart-Treves syndrome: pathogenesis and management. *J Am Acad Dermatol.* 2012;67(6):1342-1348.
19. Bechara F.G., Gambichler T., Stucker M., et al. Histomorphologic correlation with routine histology and optical coherence tomography. *Skin Res Technol.* 2004;10(3):169-173.