

## Letter to the Editor

# Two cases of lymphangioma circumscriptum successfully treated with pulsed dye laser and cryotherapy

Sir,

Lymphangiomas are congenital malformations of the lymphatic system due to hamartomatosis. They can be classified as deep (macrocytic) or superficial (microcytic) according to the depth of placement and size of the lymphatic ducts. Lymphangioma circumscriptum has been placed in the superficial subgroup under the deep (macrocytic) group.<sup>[1,2]</sup>

In lymphangioma circumscriptum, vesicles that are 1–4 mm in diameter can be clear, but depending on the amount of blood in the lymphatic ducts, they can also be pink-red in color.<sup>[1]</sup>

Various treatment options for lymphangioma circumscriptum include surgery, sclerotherapy, cryotherapy, and lasers. Although these treatment options may yield positive results in some cases, relapses are often reported due to the deep placement of the lesions.<sup>[1]</sup> Here, two cases of lymphangioma circumscriptum successfully treated with a pulsed dye laser (PDL) and cryotherapy are presented.

### Case 1

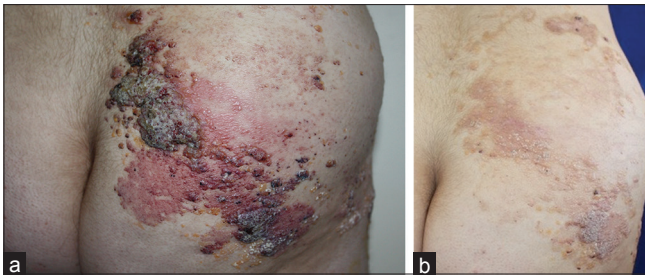
A 31-year-old female patient presented to our clinic with pink-red pimples that she had had since birth, and that were dispersed throughout the right gluteal region, down to the right femoral area, and had grown over time in both number and size. The patient reported that her lesions became infected

twice per year. Nothing in particular was noted in her family history or her laboratory tests.

The dermatological examination showed vesicle-like lesions that began in the right gluteal region and extended to the inner side of the right femoral area. These were 1–5 mm in diameter, had colors varying between yellowish pink and red, and while some had a tendency to form groups, other were dispersed throughout the area [Figure 1a and 2a]. A punch biopsy was taken from the patient, and histopathology revealed that there were many dilated lymphatic ducts and lymphocyte groups around these ducts in the superficial dermis [Figure 3]. Because some lesions were hemorrhagic and others had a translucent yellowish color, a PDL was applied for the hemorrhagic lesions and cryotherapy for the translucent yellowish ones. PDL with a cap that is 5 mm in diameter, a 350 ms pulse time, and 585 nm wave length was used on the patient's lesions. The laser treatment consisted of 5 sessions at 4-week intervals, beginning with a dose of 7 J/cm<sup>2</sup>, and in every session the dose was increased by 0.5 J/cm<sup>2</sup>, so that the final dose was 9 J/cm<sup>2</sup>. Meanwhile, cryotherapy was applied to the translucent yellowish vesicles in 5 sessions of double freeze–thaw cycles at 3-week intervals. With the combination of PDL and cryotherapy, the patient's lesions regressed [Figures 1b, and 2b]. We followed up this patient for nearly two years. Small sized new lesions measuring a few millimetres appeared during this time and we performed cryotherapy (five sessions at three month intervals). After treatment, significant improvement was observed in the patient's lesions.

### Case 2

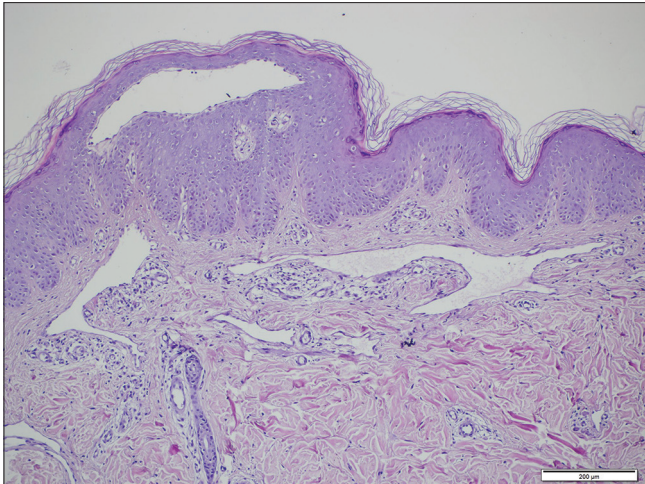
A 16-year-old female patient presented to our clinic with small blisters that had existed since she was 1 year old. Dermatological examination of the patient, whose family history and laboratory



**Figure 1:** (a) Vesicle-like, hemorrhagic, grouped lesions that began in the right gluteal area and extended to the inner side of the right femoral area. (b) Post-treatment appearance



**Figure 2:** (a) Grouped lymphangioma lesions extended to the inner side of the right femoral area. (b) Post-treatment appearance



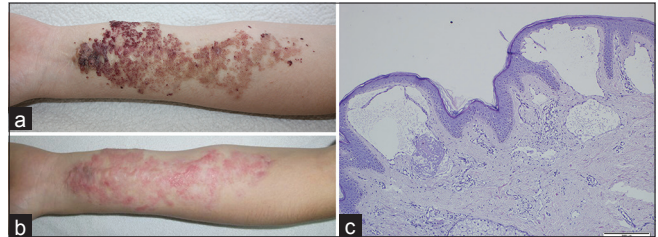
**Figure 3:** Dilated lymphatic ducts and lymphocyte groups in the superficial dermis (H and E,  $\times 100$ )

tests showed nothing significant, revealed vesicular lesions that were 1–4 mm in diameter (some in groups and some scattered), which had colors varying from yellowish pink to red. These blisters extended along on the medial side of her left forearm.

The vesicles merged distally, forming a red plaque 4  $\times$  7 cm in size [Figure 4a]. A punch biopsy was taken and revealed many dilated lymphatic ducts and lymphocyte groups around these ducts [Figure 4b]. Because the patient's lesions were mostly pink-red in color and hemorrhagic in appearance, the patient was started on PDL treatment. Laser treatment was begun at a dose of 7 J/cm<sup>2</sup> with 0.5 J/cm<sup>2</sup> increments over 5 sessions at 4-week intervals, to reach a final dose of 9 J/cm<sup>2</sup>.

After treatment, significant regression was observed in the patient's lesions, with a hemorrhagic appearance. However, some translucent yellowish lesions did not regress, so cryotherapy was applied for these lesions; after 3 sessions they completely regressed with postinflammatory hyperpigmentation [Figures 4c].

Lymphangioma circumscriptum is a benign lymphatic ectasia with two components: A clinically apparent dermal vascular component and a not so apparent deep subcutaneous cisternal



**Figure 4:** (a) Small lymphangiomas located on the arm, with colors varying from pink to dark red. (b) Post-treatment appearance. (c) Dilated lymphatic ducts and lymphocyte groups in the dermis (H and E,  $\times 100$ )

structure.<sup>[3]</sup> Although it is characterized by translucent, pink-red vesicle bundles with thin membranes sometimes filled with blood, the vesicles have clear edges, and can be scattered or in groups.<sup>[4]</sup>

For the differential diagnosis of lymphangioma circumscriptum, lymphangiectasia, hemangioma, verruca, molluscum contagiosum, angiokeratoma, and lymphangi endothelioma should be considered.<sup>[2]</sup> The risk of malignant conversion is very low for lymphangioma circumscriptum. In the literature, one case was reported to develop squamous cell carcinoma, and one developed lymphangiosarcoma.<sup>[5]</sup>

Although the main objective of the treatment of lymphangioma circumscriptum is cosmetic, treatment for constant lymphatic and blood leakage, pain, edema, and persistent infections is also relevant. Many successful treatments, including surgical excision, incision and drainage, injection of sclerosant agents, electrocautery, cryotherapy, radiotherapy, and lasers have been reported. The most common treatment with the lowest recurrence rate among these methods is surgical treatment.<sup>[6]</sup>

For the treatment of lymphangioma circumscriptum, many ablative and nonablative lasers have been used; however, there have been few reports in the literature. Although successful results have been gained with CO<sub>2</sub> lasers, it has its disadvantages, such as the requirement of anesthesia, and the risk of scarring.<sup>[7]</sup>

PDLs used for treating vascular lesions are also used for treating lymphangioma circumscriptum. PDLs, with higher pulse durations, are more consistent with the thermal relaxation time of the tissues. Therefore, PDLs carry a lower risk of complication such as pigmentation or scarring.<sup>[8]</sup> PDLs cause selective vascular damage without harming the surrounding tissues; they can be used in all lesions having a vascular component, such as warts or hypertrophic scars.<sup>[8,9]</sup> Many parameters determine the effectiveness of treatment, such as localization, depth, and hemorrhagic content of the vascular lesion.<sup>[9]</sup>

Railan *et al.* reported the treatment of two cases of lymphangioma circumscriptum with PDL; though successful results were gained, recurrence was observed in one case after 3 years.<sup>[7]</sup> We too had successful results using a combination of PDL and cryotherapy. The lesions in our patient 1 were very large plaques; therefore, we could not rely on only one method of treatment. As she had many lymphangiomas, we performed PDL on the hemorrhagic lesions, and cryotherapy on the other lesions. She responded well to a combination of PDL and cryotherapy.

Clinicians should therefore not overlook nonsurgical treatment methods such as PDL and cryotherapy in cases of lymphangioma circumscriptum, which could be both effective and recurrence-free.

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