



## Extensive scrotal lymphangioma circumscriptum: A case report with successful treatment with staged CO2 laser ablation

Kiana Saade, Harold C. Hamann<sup>\*</sup>, Sean T. McGinley, Nicole Alavi-Dunn, Nicole Belko, Teresa Danforth

Department of Urology, Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, Buffalo, NY, 14203, USA

### ARTICLE INFO

#### Keywords:

Lymphangioma circumscriptum  
Radiation therapy  
Non-venereal penoscrotal lesions  
CO2 laser ablation

### BRIEF ABSTRACT

Scrotal lymphangioma circumscriptum (LC) is an uncommon, benign skin disorder with multiple treatment modalities. We present staged treatment of extensive LC of the scrotum using staged carbon dioxide (CO2) laser ablation. We achieved near complete resolution of lesions with excellent cosmesis in the span of four months, from presentation to treatment. Therefore, the use of staged CO2 laser ablation should be strongly considered when LC presents in the genitourinary (GU) system, as it can lead to good cosmetic outcomes and prompt symptomatic relief in a short period of time.

### 1. Introduction

Lymphangioma circumscriptum (LC) is a benign skin disorder caused by malformations of the lymphatic channels, most commonly occurring on the trunk, axilla, thigh, buttock area, and oral cavity, and less commonly on the scrotum and penis.<sup>1</sup> Despite being commonly mistaken for viral warts, herpes zoster, and molluscum contagiosum, it uniquely appears as red thin-walled translucent vesicles.<sup>2</sup> Many treatment modalities are described in the literature such as Nd:YAG, diode lasers, electrocoagulation, cryotherapy and wide surgical excision. We present a case of extensive scrotal LC successfully treated with staged CO2 laser ablation.

### 2. Case presentation

A 58-year-old male with past medical history significant for hypertension, hyperlipidemia, Hodgkin's lymphoma presented with flesh-colored lesions on his scrotum without penile involvement or active signs of infection, though drainage was present (Fig. 1a). His lymphoma was treated in 1997 with external beam radiation therapy to chest and neck with subsequent recurrence in 1999 to his groin that was treated with local radiation and chemotherapy. As these lesions did not appear typical of an infection or known benign lesions, the decision was made to biopsy these lesions. Excisional biopsy was carried out with pathology

noting dilated lymphovascular channels highlighted by D-240, CD31 and CD34 immunostains (Fig. 1b). Patchy, chronic dermal inflammation and focal surface ulceration was also noted. These findings were consistent with the diagnosis of LC. Two months after the biopsy, he underwent staged CO2 laser ablation of the scrotal lesions. By targeting the deep lymphovascular channels, all visible and palpable lesions were ablated successfully. Collagenase was subsequently applied. The patient was seen in clinic two months post-operatively and was noted to have decreased lesion burden with successful treatment in the treated areas. (Fig. 1c). Approximately 1.5 months after the first CO2 laser ablation, the patient was taken back for another CO2 ablation. The patient presented one month later for post-operative clinic appointment, which showed near complete resolution of lesions with excellent cosmesis (Fig. 1d). The total elapsed time between diagnosis and final treatment was approximately four months. Follow up three months after the patient's last surgery did not reveal any recurrence of these lesions.

### 3. Discussion

LC, also known as microcystic lymphatic malformation, can be congenital or due to secondary causes such as radiotherapy, infections, or surgical procedures.<sup>3</sup> While rare in general, it is even less commonly seen in the genitalia, with only 8 cases of penile LC reported between 1947 and 2018.<sup>1</sup> As with our patient, they typically appear as multiple

*Abbreviations:* LC, Lymphangioma circumscriptum; CO2, Carbon dioxide; GU, Genitourinary.

<sup>\*</sup> Corresponding author. Department of Urology Jacobs School of Medicine and Biomedical Sciences, University at Buffalo, 100 High Street, B280, Buffalo, NY, 14203, USA.

*E-mail address:* [hhamann@buffalo.edu](mailto:hhamann@buffalo.edu) (H.C. Hamann).

<https://doi.org/10.1016/j.eucr.2021.101871>

Received 9 August 2021; Received in revised form 16 September 2021; Accepted 27 September 2021

Available online 27 September 2021

2214-4420/© 2021 The Authors.

Published by Elsevier Inc.

This is an open access article under the CC BY-NC-ND license

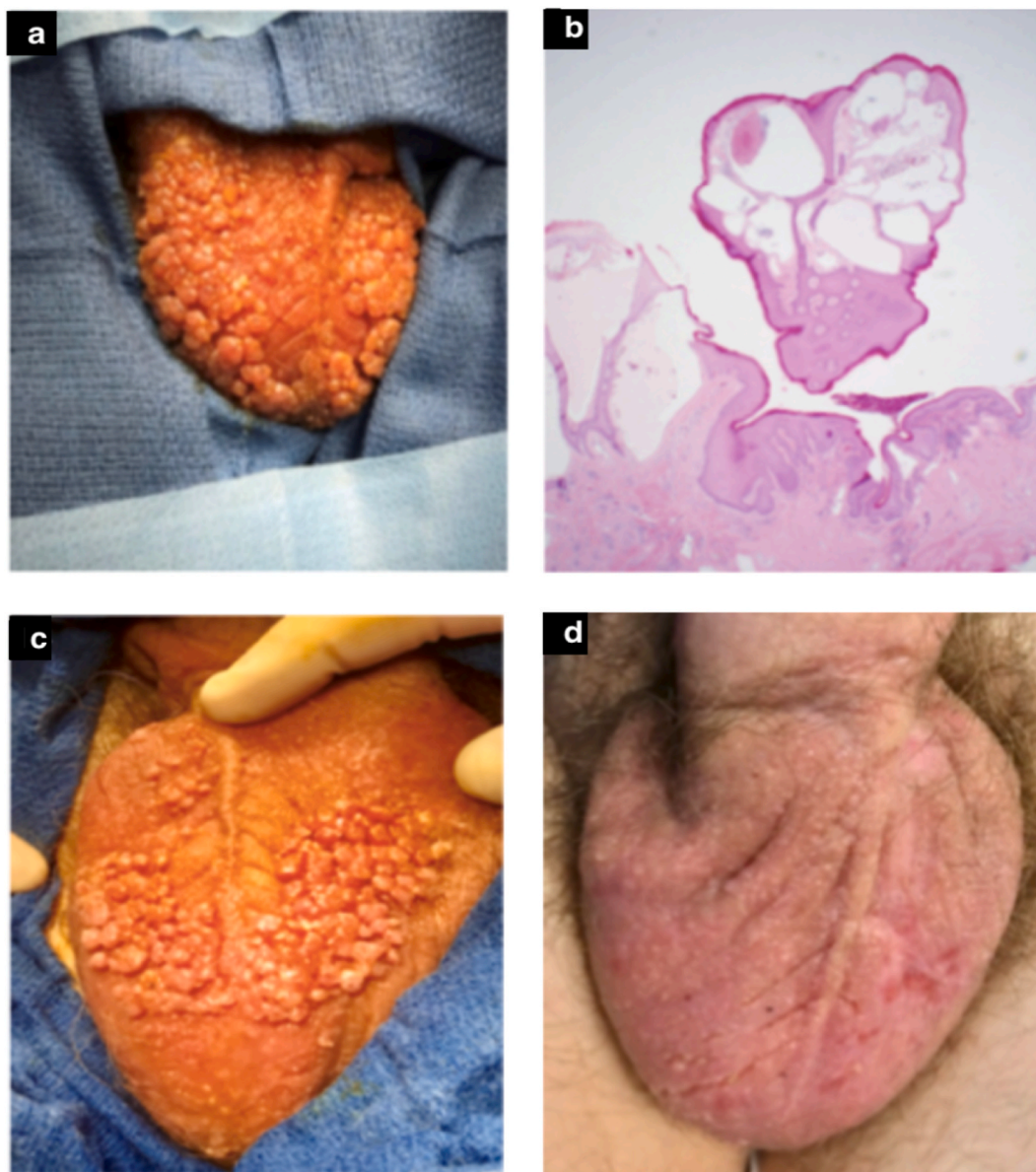
(<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

scattered or grouped translucent vesicles. With a high index of suspicion, an excisional biopsy should be performed. On histology, the diagnosis is confirmed by seeing dilated lymphatic channels with inflammatory cells in their lumina are seen in the subepithelium.<sup>2</sup>

While surgical excision is still considered the gold standard of treatment, extensive disease could necessitate reconstruction. Our case demonstrates successful symptomatic resolution of scrotal LC using two-session staged CO2 laser ablation. In contrast to previously reported successful ablation of these lesions with CO2 lasers, we felt a staged approach would offer the least post-operative pain and maximize the overall cosmetic appearance given that the LC involved the entire scrotum. In a similar case, Schulz et al. used a 4-session staged CO2 laser ablation approach.<sup>3</sup> However, they reported that their patient healed with white scar-like lesions, whereas our patient healed with flesh-colored scars, possibly due to fewer sessions and less exposure to CO2 laser ablation. We attribute this different cosmetic outcome to not only our staged process of ablation, but also to the depth of penetration of the laser. Since the pathology of LC stems from the clinically obvious

dermal component and the abnormal subcutaneous lymphatic cisternae, it is important to focus energy on the subdermal component of the LC as well. Not only did we erupt the dermal vesicles, but we applied the laser on the entrance to the cisternae for up to 45 seconds. Since the lymphatic cistern also derives fluid from surrounding tissue, the laser was also focused obliquely on the few millimeters around each ruptured vesicle.

Other non-invasive treatment methods have been described in the literature, including Nd:YAG, diode lasers, electrocoagulation, and cryotherapy. In Sasaki et al.'s report, Nd:YAG was used in combination with CO2 therapy,<sup>4</sup> with Nd:YAG being preferred by the patient. Unlike CO2 laser ablation, Nd:YAG stopped lymphatic fluid discharge. On the second session of the Nd:YAG however, the patient developed a temporary 4mm linear scar. Compared to our technique, the Nd:YAG required four sessions, once monthly, while ours was completed in two sessions in one and a half months. Finally, while excisional surgery may be considered the "gold standard," it is still a more invasive technique that may result in complications including risk of recurrence secondary to incomplete resection.<sup>5</sup>



**Fig. 1.** Clinical images showing (a) lymphangioma circumscriptum (LC) of the scrotum prior to treatment with CO2 ablation, (b) histologic image from scrotal biopsy demonstrating dilated lymphatic channels of the dermis and hyperkeratosis of epidermis (HE, x2), (c) LC of the scrotum two months after first laser ablation, (d) LC of the scrotum one month after second laser ablation.

#### 4. Conclusion

Cutaneous lesions are not encountered as ubiquitously in a urologist's training, thus learning facets of urologic dermatology are gained mostly through experience. Given the nonspecific presenting symptoms such as swelling, oozing, pain, and infection, LC, on the genitalia, may be confused with herpetic lesions or genital warts. With a preponderance of GU cancers being treated with lymph node dissections or radiation therapy, LC should be considered in the differential diagnosis of cutaneous lesions of the external genitalia that appear post-therapy. When in doubt, a biopsy of the lesion in question should be performed prior to consideration of therapy.

In summary, recognition and pathological identification of LC is crucial to its treatment. Once identified, the use of staged CO<sub>2</sub> laser ablation should be strongly considered when LC presents in the GU system as it can lead to good cosmetic outcomes and prompt symptomatic relief in a short period of time.

#### Funding

This research did not receive any specific grant from funding

agencies in the public, commercial, or not-for-profit sectors.

#### Declaration of competing interest

The authors have no competing interests to declare.

#### References

1. Bonini J, Ducharme O, Ponroy B, Dauendorffer JN, Baubion E. Acquired lymphangioma circumscriptum of the penis treated by electrocoagulation. *Case Rep Dermatol.* 2019;11(3):260–263. <https://doi.org/10.1159/000503137>. Published 2019 Sep. 30.
2. Fatima S, Uddin N, Idrees R, et al. Lymphangioma circumscriptum: clinicopathological spectrum of 29 cases. *J Coll Physicians Surg Pak.* 2015;25(9):658–661.
3. Schulz D, Lein A, Nicula AP, Schierle K, Solovan C. Lymphangioma circumscriptum post radiotherapy for penile cancer treated with CO<sub>2</sub> laser. *Acta Dermatovenerol Croat.* 2018;26(1):53–57.
4. Sasaki R, Negishi K, Akita H, Suzuki K, Matsunaga K. Successful treatment of congenital lymphangioma circumscriptum of the vulva with CO<sub>2</sub> and long-pulsed Nd:YAG lasers. *Case Rep Dermatol.* 2014;6(1):1–4. <https://doi.org/10.1159/000358006>. Published 2014 Jan 8.
5. Haroon S, Hasan SH. Lymphangioma circumscriptum in the scrotum: a case report. *J Med Case Rep.* 2012;6:233. <https://doi.org/10.1186/1752-1947-6-233>. Published 2012 Aug 9.