

Clinical Spectrum and Therapeutic Approaches in Lymphangioma Circumscriptum: A Case Series of Nine Patients

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ABSTRACT

Lymphangioma Circumscriptum (LC) is a rare hamartomatous condition affecting the lymphatic system. It is also referred to as microcystic lymphatic malformation. Treating this condition is both intriguing and challenging. The present case series is about nine patients (7 males, 2 females) with LC presenting at different locations, highlighting their clinical, dermoscopic, histopathological and different management strategies. The patients ranged in age from seven to 58 years and included both congenital and acquired forms of LC. Clinically, the lesions were slow growing, asymptomatic, and grouped, involves sites such as the beard area, vulva, neck, abdomen, gluteal region, and distal extremities. On Dermoscopy examination, pale yellow or translucent lacunae separated by septa, with occasional hypopyon like features due to blood sedimentation were noted. Histopathological examination revealed characteristic thin walled lymphatic channels in superficial dermis. To rule out deeper involvement Imaging was utilised selectively. Various treatment modalities were employed, including Carbon dioxide (CO₂) laser, Radiofrequency Ablation (RFA), cryotherapy Pulsed Dye Laser (PDL), intralesional bleomycin, and combination therapies, tailored according to lesion morphology and depth. Most patients achieved satisfactory clearance with minimal adverse effects, and no recurrence were noted during follow-up. LC, though benign, poses aesthetic, psychological, and therapeutic concerns, especially when occurring at atypical sites. Even though surgical excision of deep lymphatic system is the mainstay treatment according to Whimster's hypothesis, this may not be feasible in patients with atypical sites and extensive involvement. In such cases minimally invasive modalities provide effective alternatives to surgery.

Keywords: Dermoscopy, Hamartomas, Lymphatic malformations

INTRODUCTION

In 1879, Fox and Fox identified lymphangioma as a hamartomatous entity, as "lymphangiectodes." Later, Malcolm Morris coined the phrase "circumscriptum" for the first time in 1889 [1]. A fascinating and uncommon dermatological disorder called LC is characterised by the presence of many or few, clustered or scattered, transparent or haemorrhagic vesicular papules that resemble frog spawn. This condition can be congenital or acquired. Congenital LC can occur as a result of malformation in the lymphatic channels. In contrast, acquired LC involves disturbances in the lymphatic system, commonly attributed to radiation therapy, surgical procedures, or infections, causing damage or obstruction to the lymphatic channels. LC is predominantly seen in paediatric populations, often involving the head and neck areas [2]. There is a notable deficiency in the reporting of detailed case series that outline the presentation, signs and treatment of this unique clinical condition. The goal of the present case series on LC is to fill the gap in the understanding of various patient histories, clinical presentations, diagnostic techniques, and treatment modalities.

CASE SERIES

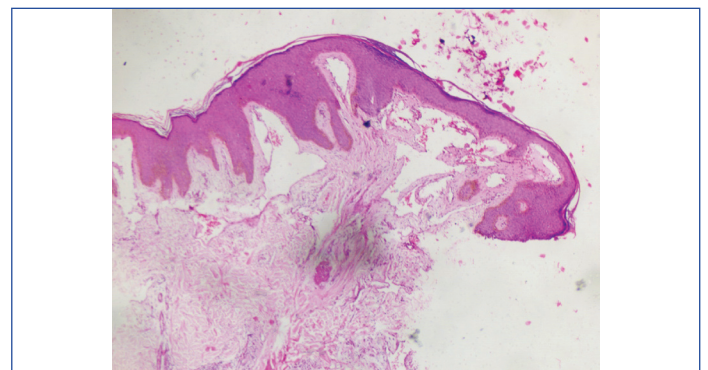
Case 1

A 16-year-old boy visited our Outpatient Department (OPD) with complaints of multiple painless, grouped, fluid-filled lesions on the right side of the abdomen for six months. On examination, multiple grouped vesicles and bullae of varying sizes were noted over the right lumbar region [Table/Fig-1]. The patient had a history of similar lesions five years ago, for which he underwent surgical excision. All routine blood investigations were normal, showing no peripheral eosinophilia with negative serology. Deeper systemic involvement was ruled out using ultrasonography. Differential diagnosis of Herpes zoster, LC and haemangioma made. Skin biopsy done



[Table/Fig-1]: Multiple grouped vesicles and bullae of varying sizes were noted over the right lumbar region.

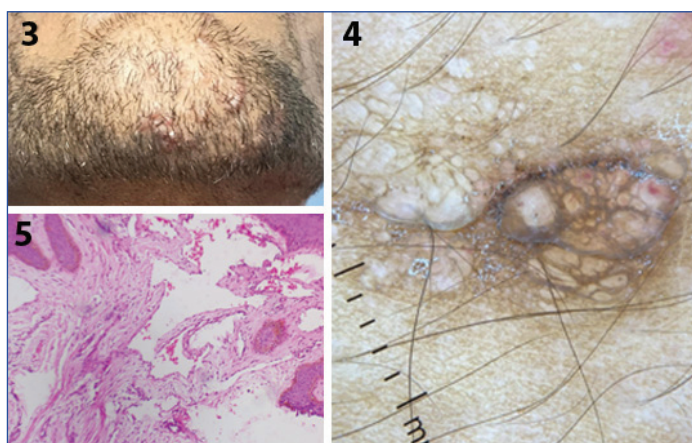
which showed dilated lymphatic channels beneath the epidermis [Table/Fig-2], hence definitive diagnosis of LC made. RFA was performed in multiple sessions, resulting in clearance. The patient was regularly followed-up every fortnightly for three months, and he did not complain of recurrence.



[Table/Fig-2]: Histopathology showing dilated lymphatic channels (Haematoxylin & Eosin 40x).

Case 2

A 28-year-old male presented with complaints of multiple grouped fluid-filled lesions over the chin area of his beard for two months [Table/Fig-3]. Patient had no associated symptom or positive family history. Dermoscopy revealed multiple flesh-coloured papules with considerable translucent lacunae and a yellow hue; a few nodules showed linear vessels [Table/Fig-4]. Skin biopsy was performed, and histopathology showed significant dilated, large lymphatic vessels in the upper dermis [Table/Fig-5], which led to a final diagnosis of LC. He was subjected to radiofrequency cauterisation of the lesions, and bleomycin injection was administered for two lesions. The lesions ulcerated after five days, and they healed with hyperpigmentation within two weeks. The patient achieved clearance and was prescribed a depigmenting cream for the pigmented area. He was under follow-up for six months and did not report recurrence.



[Table/Fig-3]: Few grouped vesicles are noted over the chin, some showing haemorrhagic crusts. **[Table/Fig-4]:** Dermoscopy showing lacunar areas surrounded by pale septa. **[Table/Fig-5]:** Dilated lymphatic spaces in the papillary and reticular dermis (Haematoxylin & Eosin 200x). (Images from left to right)

Case 3

A 58-year-old female, housewife by occupation, presented with numerous raised lesions on the genitalia for two years. She had a history of cervical cancer for 10 years, for which surgery and radiotherapy were done. There were no similar cases reported in the family. On examination, multiple, grouped papules and vesicles, which resemble “Frog spawn”, were noted over the bilateral vulva [Table/Fig-6]. Dermoscopy revealed multiple flesh-coloured papules along with translucent lacunae and a yellow hue, and a few nodules showing linear vessels [Table/Fig-7]. Differential diagnosis of cutaneous LC included haemangioma, angiokeratoma, molluscum contagiosum, and warts. All routine blood investigations were normal, and no peripheral eosinophilia was observed. She did not have any significant past medical history of sexually transmitted diseases, and the serology was negative. A skin biopsy was performed, and histopathology revealed thin-walled, dilated lymph channels beneath the epidermis, without endothelial swelling. Few inflammatory infiltrates were noted in the dermis of the section examined, leading to a diagnosis of LC [Table/Fig-8]. She underwent multiple sessions of radiofrequency treatment, and clearance was achieved and patient was followed for one year with no recurrence.

Case 4

A 28-year-old female with skin coloured raised lesions over the neck for four months [Table/Fig-9]. There was no family history or history of any trauma. The patient did not agree to a skin biopsy. Dermoscopy showed hypopyon like lesions [Table/Fig-10]. Clinical diagnosis of LC was made with the help of dermoscopy. The patient underwent surgical radiofrequency treatment, and clearance of the lesions was achieved with no recurrence.



[Table/Fig-6]: Multiple grouped papular lesions on the bilateral vulva. **[Table/Fig-7]** Dermoscopy showing multiple flesh-coloured papules along with translucent red lacunae. **[Table/Fig-8]** Section of the specimen showing features of Lymphangioma Circumscriptum (LC) with acanthosis, hyperkeratosis of the epidermis with dilated thin lymphatic channels in the upper dermis and eosinophilic protein material in the papillary dermis. (Haematoxylin & Eosin, 40x). (Images from left to right)



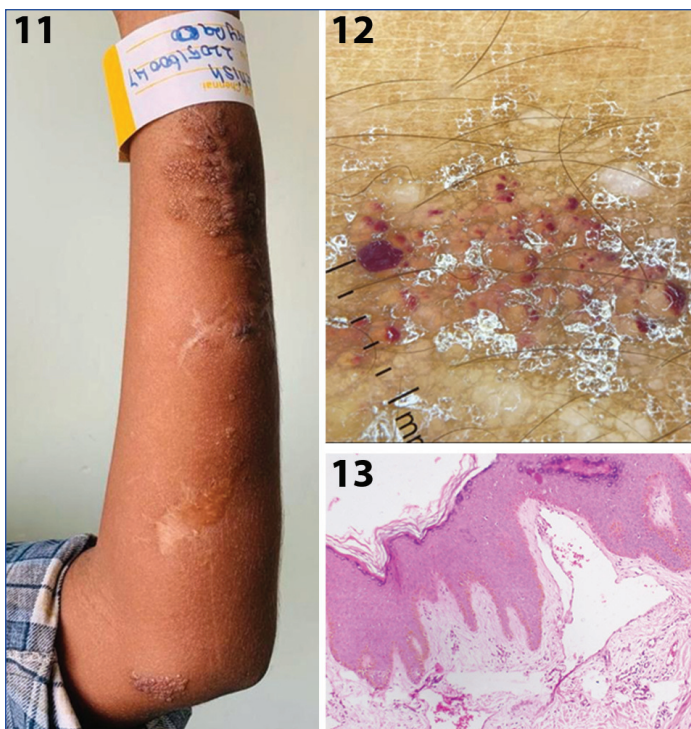
[Table/Fig-9]: Multiple skin coloured papules in the neck. **[Table/Fig-10]** Dermoscopy showing hypopyon like lesions. (Images from left to right)

Case 5

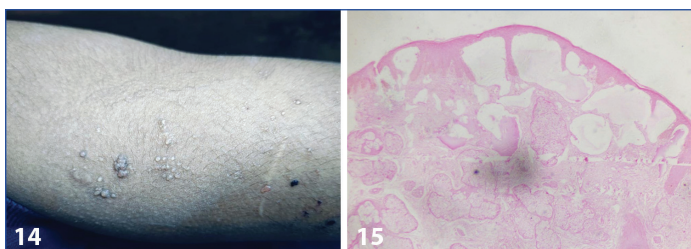
A 31-year-old male with complaints of asymptomatic grouped pale-coloured raised lesions near the right elbow and the right wrist for four months. On examination, multiple grouped skin-coloured papules and a few vesicles were noted near the elbow and the volar aspect of the right forearm [Table/Fig-11]. On dermoscopic examination, it showed multiple pale yellowish, well-circumscribed lacunae surrounded by pale septa with few containing blood, which typically accumulated in the lower part of the lacuna [Table/Fig-12]. The biopsy results were consistent with the features of LC [Table/Fig-13]. Different treatment modalities were employed for the lesions. For haemorrhagic lesions, PDL therapy was applied, while cryotherapy was used for translucent yellowish ones. The lesions improved significantly, and follow-up was conducted over a period of nearly two years and no recurrence of lesion was noted.

Case 6

An 18-year-old male presented to our skin OPD with complaints of asymptomatic, multiple, filled, raised lesions over the right arm for the past one year [Table/Fig-14]. On examination, skin coloured grouped papules and vesicles were noted over the extensor aspect of the right arm. Clinical diagnosis of LC was made and confirmed by skin biopsy which revealed dilated lymphatic channels with eosinophilic proteinaceous material in the superficial dermis [Table/Fig-15]. Cryotherapy and radiofrequency removal of the lesions was done and achieved complete clearance with no further recurrence.



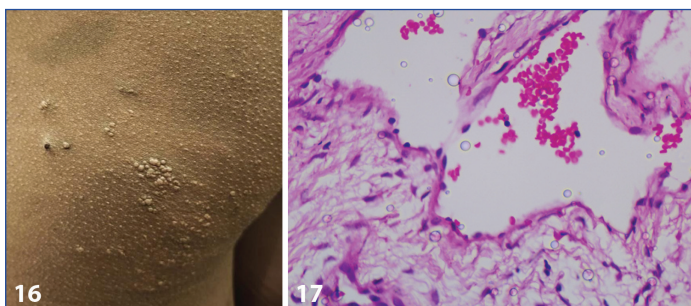
[Table/Fig-11]: Multiple grouped papules coalescing to form plaques on the extensor surface of the forearm and the elbow. **[Table/Fig-12]** Dermoscopy of the lesion showing reddish lacunae and the "hypopyon" like features. **[Table/Fig-13]** Histopathology showing dilated lymphatic channels in the papillary dermis. (Haematoxylin & Eosin, 40x). (Images from left to right)



[Table/Fig-14]: Multiple grouped vesicles noted over the extensor aspect of the right arm. **[Table/Fig-15]** Histopathology showing basket weave orthokeratosis with multiple lymphatic spaces in the dermis. (H&E,40x). (Images from left to right)

Case 7

A seven-year-old male child was brought to our outpatient department with complaints of multiple fluid-filled, raised skin lesions over the left gluteal region for the past six months [Table/Fig-16]. The patient is a diagnosed case of lymphangioma of the left gluteal region, as confirmed by Magnetic Resonance Imaging (MRI) three years ago, and was on oral sirolimus for two months, discontinued after two months due to allergic reactions, and underwent sclerotherapy. There were no similar complaints among the family members present. On examination, a few discrete and grouped vesicles were noted over the left gluteal region, dermoscopy showed hypopyon like phenomenon, skin biopsy revealed dilated lymphatic channels in the dermis [Table/Fig-17] and diagnosed as a case of LC. Patient underwent two



[Table/Fig-16]: Multiple discrete and grouped vesicles over left gluteal region. **[Table/Fig-17]** Histopathology showing dilated lymphatic channels (Haematoxylin & Eosin 400x). (Images from left to right)

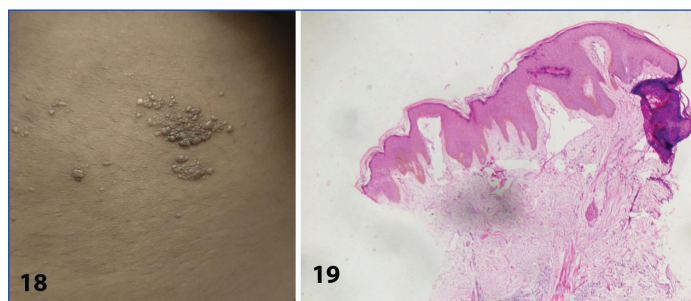
sessions of radiofrequency removal and is on regular follow-up with no recurrence.

Case 8

A 25-year-old male patient presented with skin coloured, asymptomatic, raised, grouped lesions over the abdomen for the past four years, which gradually increased in size and number [Table/Fig-18]. Skin biopsy was done, and histopathology revealed numerous dilated lymphatic spaces in superficial dermis with perivascular lymphocytic infiltration [Table/Fig-19], which led to the diagnosis of LC. Ablative CO₂ laser was done, and the patient obtained complete clearance. Patient was followed-up for six months with no recurrence of the lesion.

Case 9

A 27-year-old male has had multiple raised, painless skin lesions over the left thigh for the past four years [Table/Fig-20]. On examination, multiple discrete and grouped vesicles were noted over the medial aspect of the left thigh, and a clinical diagnosis of LC was made. The patient was advised about different modalities of treatment available but he was not willing for treatment at present.



[Table/Fig-18]: Skin coloured multiple papules coalescing to form plaque noted over the left lumbar region. **[Table/Fig-19]** Histopathology showing dilated lymphatic vessels with perivascular infiltration (Haematoxylin & Eosin 40x). (Images from left to right)



[Table/Fig-20]: Multiple grouped vesicles over medial aspect of left thigh.

A summary of all the nine cases have been presented in [Table/Fig-21].

DISCUSSION

A rare hamartomatous condition, LC, accounts for approximately 4% of all vascular malformations and 25% of benign vascular tumours [3]. The common sites of involvement in the present case series include the head and neck (22%), upper limbs (22%), and lower limbs (22%), trunk (22%) and anogenital region (11%). However, Fatima S et al., reported that the anogenital region (24%), followed by the extremities (17%) and the tongue (14%), was the most common site in their cohort of Pakistani patients [3].

All nine patients in the study developed lesions later in life (acquired), and none were congenital. Acquired LC in adults can develop de novo without clear precipitating factors or may develop at sites of prior

Case	Age/ sex	Site	Clinical features	Investigation	Treatment
1	16/M	Right lumbar region	Asymptomatic multiple grouped vesicles	Skin biopsy	Radiofrequency Ablation (RFA)
2	28/M	Chin	Painless grouped papules.	Dermoscopy, Skin biopsy	Injection bleomycin, Radio frequency cauterisation.
3	58/F	Vulva	Multiple grouped papules, vesicles	Dermoscopy, Skin biopsy	management as Radiofrequency ablation
4	28/F	Right-side of neck	Asymptomatic discrete vesicles	Dermoscopy	Radiofrequency cauterisation.
5	31/M	Right elbow and wrist	Asymptomatic grouped papules and plaques	Dermoscopy, skin biopsy	Cryotherapy, Pulsed dye laser
6	18/M	Right arm	Painless grouped papules and vesicles	Skin biopsy	Cryotherapy and Radiofrequency
7	7/ M	Left gluteal region	Multiple discrete, grouped vesicles	MRI, dermoscopy, skin biopsy	Surgical management
8	25/M	Abdomen	Asymptomatic grouped vesicles	Skin biopsy	Ablative CO ₂ laser
9	27/M	Left thigh	Multiple discrete and grouped papules	Clinical diagnosis	Conservative management

[Table/Fig-21]: Characteristics of patients with Lymphangioma Circumscriptum (LC).

surgical excision, radiation therapy for malignancies, pelvic surgery, and inflammatory diseases [4]. Almalki MK et al., (2024) documented a case of scrotal LC in 35-year-old male with no previous exposure to infections, trauma, surgery, or radiation, as noted in eight out of nine cases in the present study [5]. The present case series encountered a single case of a 58-year-old female patient with cervical cancer who developed vulvar LC following surgery and radiotherapy. Valente K et al., (2016) reported a similar case of acquired vulvar LC in a 55-year-old woman following chemotherapy, radiation, and brachytherapy for cervical cancer [6]. Jappe U et al., (2002) described LC of the vulva following surgery and radiotherapy of cervical cancer, proposing that lymphatic damage from radiation combined with lymphoedema creates conditions conducive to lymphatic vessel proliferation and ectasia [7].

Ages of the patients ranged from 7 to 58 years (mean age 25.1 years), with a male-to-female ratio of 2:1. This was in concordance with Fatima S et al., (2015), who reported a series of 29 patients with a mean age of 27.17±15.5 years, with male predominance (62%), which closely parallels our findings [3].

Clinically, LC is characterised by the appearance of small, transparent vesicles with thin walls, measuring 1-4 mm, and containing crystalline or serous fluid, which occasionally may be haemorrhagic. One patient in the study had haemorrhagic vesicles within the LC lesions. Similar haemorrhagic lesions were reported by Shreshta S et al., (2020) and Alrashdan MS et al., (2018) and pose a diagnostic challenge, mimicking angiokeratomas or other vascular lesions and presenting with pain and bleeding from the lesions [8,9].

Most patients were asymptomatic, except for one who experienced vulvar itching during the study. This is consistent with studies by Patel GA et al., (2009) and Sinha A et al., (2015), who reported that most LC lesions were asymptomatic, with patients primarily seeking medical attention for cosmetic concerns or incidental discovery during routine examination [10,11]. Dermoscopy and biopsy can be employed as necessary to confirm the diagnosis [12]. The characteristic dermoscopic features observed are reddish lacunae and hypopyon-like features, which are well-established hallmarks of LC, and these typical patterns were observed in almost all patients in whom dermoscopy was performed [13]. A study by Zaballos P et al., (2018) in 45 patients found that the most common structure in dermoscopy was the presence of lacunae (89% of cases) [14].

Skin biopsy remains the gold standard for definitive diagnosis [12,15]. Characteristic microscopic findings include dilated, thin-walled lymphatic vessels in the papillary dermis and subcutaneous tissue, lined by flattened endothelial cells that often contain lymphatic fluid and erythrocytes, findings consistent with those reported in the present study. Although immunohistochemistry was not performed due to cost constraints, several studies have emphasised the value of lymphatic endothelial markers, such as CD31 and D2-40 (podoplanin), in this context [6,16]. Lu L et al., demonstrated that CD31-positive staining and cystic-dilated spaces with flattened endothelia are diagnostic features of LC [16].

In the current study, imaging was either not required or the patient declined to undergo imaging. MRI remains the gold standard for determining the full extent of lesions, particularly their deeper components, and is essential for surgical planning, as highlighted by Shrestha S et al., and Gomides MDA et al., [8,15].

The RFA was used in three patients in the study who responded well to the procedure, as previously reported by Omprakash HM in 2008 [17]. Niti K et al., (2010) also combined RFA with sclerotherapy to achieve near-complete clearance in nine of 10 patients; RFA ablates lesions and achieves haemostasis, while the sclerosant reaches deeper vascular lesions to prevent recurrence [18]. Ablative CO₂ laser treatment was performed in a single patient, enabling precise vaporisation with excellent haemostasis. Savas JA et al., (2013) conducted a systematic review that identified 16 studies involving 28 patients treated with CO₂ laser for LC [19]. Karadag AS et al., (2015) also reported two cases of LC successfully treated with PDL and cryotherapy, with a lower risk of complications, such as pigmentation or scarring, compared with CO₂ laser. Cryotherapy alone is effective as adjunctive or monotherapy, particularly for smaller lesions [20]. The use of intralesional bleomycin for the 28-year-old male patient with chin lesions was supported by Khurana A et al., (2018), who reported successful treatment of extensive LC of the flank with a combination of intralesional bleomycin and RFA, achieving near-complete resolution with no significant recurrence [21]. Immunomodulators such as topical imiquimod (5% cream), oral and topical Sirolimus (an mTOR inhibitor) have demonstrated efficacy in some studies [22,23]. Surgical excision, performed in two of our cases, remains the most definitive therapeutic approach, with recurrence rates of 17-23% when complete removal of deep dermal and subcutaneous components is achieved [24]. Recurrence of LC was noted in the seven-year-old boy with lesions involving the thigh and gluteal region after surgical excision of the lesions, within six months.

CONCLUSION(S)

The present case series highlights the evolving approach to LC management. In extensive and invasive LC of the trunk or proximal extremity, surgical excision remains the mainstay, where complete removal may be difficult, and recurrence is common if deep components remain. Superficial RFA is safe and economical in patients with higher surgical risk and may be beneficial in cases with localised genital or perigenital LC (vulva, scrotum, perineum) where wide excision and scarring would distort anatomy or impair sexual/urinary function. The major drawback of RF treatment is that deeper cisterns may persist; long-term recurrence beyond 1-2 years remains unknown. On the other hand, CO₂, PDL, and (long-pulsed) Neodymium-doped Yttrium Aluminium Garnet laser (Nd:YAG) lasers may be preferred when precise ablation of superficial vesicles with a good cosmetic outcome is paramount, particularly in genital and mucosal LC. However, cost and availability limit its widespread use in most settings.

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