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## Dermatology Online Journal

### Title

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### Permalink

<https://escholarship.org/uc/item/9pr342qv>

### Journal

Dermatology Online Journal, 24(8)

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### Publication Date

2018

### DOI

10.5070/D3248041141

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Peer reviewed

# Lymphangioma circumscriptum of glans penis: a report of two cases

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## Abstract

Lymphangioma circumscriptum is a developmental anomaly of lymphatic vessels, which appear as aggregates of clear or hemorrhagic vesicles on the skin or mouth. Glans penis involvement is very uncommon. Because of the sensitivity of the area, possible functional, cosmetic, or psychologic disturbances can result. Lymphangioma circumscriptum is rarely found on this location; hence, vigilance and awareness of this entity is necessary for a swift and proper diagnosis. Two cases are presented on the account of their rarity and unique representation.

*Keywords: lymphangioma circumscriptum, glans penis, lymphatic anomaly*

## Introduction

Lymphangioma circumscriptum (LC) is a lymphatic malformation that usually presents in infancy and childhood [1], but can also be acquired. It sometimes develops in young adults and more rarely, in older individuals. One of the less frequent sites of involvement is the glans penis. Up to date there are very few reports of penile lesions in the English literature [2-10]. Herein, we present two cases of LC, the first for its unusual site and occurrence in an older age, the second for its unique presentation of a single black papule on the tip of the glans penis.

## Case Synopsis

### Case 1

A 49-year-old man presented with a 5-month history of linear purple hemorrhagic papulovesicles on his glans penis (Figure 1). After an initial period of growth there was no further expansion of the lesions. The patient had no pain or pruritus but complained of intermittent swelling of the papules. There was no previous infection, trauma, or surgical procedure and he had not undergone any kind of treatment or diagnostic measures. We obtained a punch biopsy from one of the papulovesicles and submitted it for histologic evaluation. The biopsy specimen revealed skin tissue with thin walled lymphatic channels, composed of single bland looking endothelial cells containing fibrin and eosinophilic material (Figure 2). There was clear fluid within lymphatic lumina and scattered lymphocytes in the stroma. The overlying epidermis was rather acanthotic.



Figure 1. Linear hemorrhagic blue-black papulovesicles across corona of glans penis.



Figure 2. Lymphatic channels are lined by single bland looking endothelial cells.

## Case 2

Our second patient was a 19-year-old man who presented to us with an asymptomatic 1.5mm black papule on the tip of the glans penis next to the urinary meatus (Figure 3). He noticed its presence for several months and was anxious about its possible malignant nature. Because of its small size we were able to completely remove it with a punch biopsy. The specimen was subsequently sent for histologic assessment. It showed similar histologic findings as in case 1, including superficial dilated lymphatic vessels located in the papillary dermis (Figure 4).

## Case Discussion

Vascular malformations can be subcategorized in terms of the principal abnormal vessels as follows; capillary malformation, venous malformation, arteriovenous malformation, and lymphatic malformation (LM). LMs are divided into macrocystic (cystic hygroma), microcystic (lymphangioma circumscriptum), and combined micro- and macrocystic LMs [11]. Most LMs are diagnosed

during infancy or childhood, although some can manifest in early and late adulthood. Vascular malformations usually do not show increased endothelial proliferation; the same is true for LMs. Hence, “lymphangioma” circumscriptum is a misnomer [1] for this subgroup of LMs (i.e. microcystic LMs).

LC appears as crops of papulovesicles filled with clear or serosanguinous fluid resembling frogspawn. Blue-black color owing to accumulation of blood is another frequent manifestation. Other signs and symptoms may include intermittent swelling, leakage of clear or bloody lymph (lymphorrhea), and infections (cellulitis). Sometimes the surface of lesions acquire a papillomatous appearance, which may be mistaken for warts [2, 8]. Molluscum contagiosum-like lesions on the penis has also been reported [2, 7]. Although virtually any part of the skin may be affected, proximal limbs, chest, axillary folds, shoulders, flanks, and perineum are sites of predilection. Male genital involvement usually manifests on the scrotum; penile lesions, especially on glans penis [3, 7] are very rare.

Lymphangioma on the penis could be acquired and appear as a result of lymphatic vessel occlusion related to infections such as frequent cellulitis [5]. Alternatively, this entity may be congenital as reported by Handa et al. [9]. However, lesions in both our cases appeared spontaneously in adulthood,



Figure 3. A jet-black papule on the tip of glans penis adjacent to the urethral orifice.

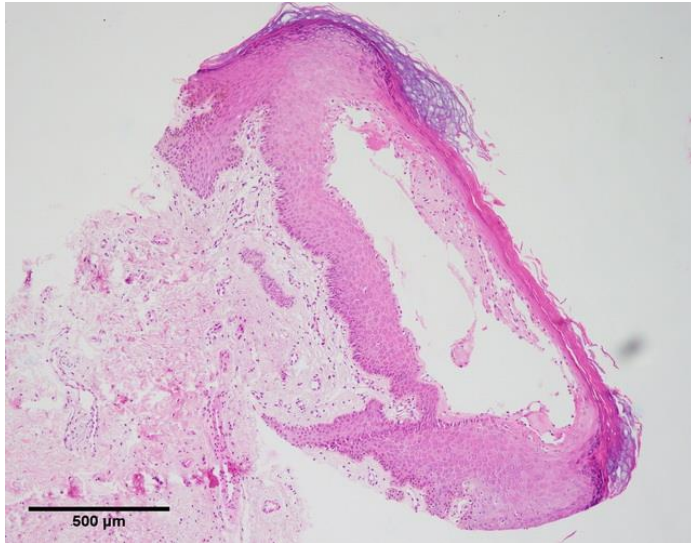


Figure 4. Dilated lymphatic vessel with clear fluid is present in the dermis.

without any history of previous infection, irradiation, or surgery. Distribution of lesions in our first patient was in the form of linear hemorrhagic blue-black papulovesicles across the corona of the glans penis. Recent onset of lesions in his late forties, its unusual location, and vascular appearance prompted us to perform a biopsy to exclude more serious conditions like Kaposi sarcoma.

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Acquired LC with a single lesion on the male genitalia is extremely rare. Adikari et al. described a 47-year-old man with a single lesion on his penis resembling a genital wart [8], whereas our second patient presented with a single jet-black papule on the tip of the penis resembling a lesion of melanocytic origin. To the best of our knowledge this is the first case of LC with this kind of unusual and unique appearance. Our first patient did not return for follow-up and the second patient did not need any further treatment, because we were able to remove the entire LC with a punch biopsy.

## Conclusion

The emergence of lesions on the glans penis can be very disturbing for patients as they may be cosmetically embarrassing, mistaken for STDs, interfere with sexual function, or misdiagnosed as a melanocytic lesion. Therefore, awareness of this rare disorder and diagnostic vigilance is required for a swift and proper diagnosis and subsequent management.