Single Case

Acquired Lymphangioma Circumscriptum of the Penis Treated by Electrocoagulation

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Abstract
Lymphangioma circumscriptum (LC) is a vascular malformation resulting from a developmental anomaly of the superficial lymphatic system of the skin. It is benign albeit uncommon. LC less frequently occurs on the penis. LC may be either congenital or acquired. Acquired cases appear to develop most frequently after interventions in the area or underlying pathologies. It is often mistaken for genital warts or molluscum contagiosum. We report here about a case of LC misdiagnosed with genital warts for 15 years. A biopsy eventually provided histopathological evidence. Various treatments are available for LC including surgical excision (which is the gold standard), CO₂ laser ablation, cryotherapy, flash lamp pulsed dye laser, and electrocoagulation therapy. For our patient, one session of electrocoagulation was performed under local anesthesia. This treatment allowed an almost complete regression of the lesion without recurrence after 3 years of follow-up. Electrocoagulation may be an efficient treatment for LC in locations that may be surgically challenging such as penis.
Introduction

Acquired lymphangioma circumscriptum (LC) is an uncommon and benign pathology. LC is a vascular malformation resulting from a developmental anomaly of the superficial lymphatic system of the skin. It rarely occurs on the penis. We report here about a case of a 69-year-old man who presented with several lesions of acquired LC on the penis. Reporting this case may increase awareness of this rare condition mimicking sexually transmitted infections, in the hope of reducing erroneous diagnosis and treatments. Surgical removal is the treatment of choice, but we demonstrate here efficient treatment with electrocoagulation.

Case Report/Case Presentation

A 69-year-old male with no other previous medical history was seen in the dermatological ward for lesions of the penis. At the clinical examination, multiple translucent vesicles of the penile shaft and glans without any associated symptoms were seen (Fig. 1). These lesions were known by the patient for 15 years. He had received neither surgery nor radiotherapy in the pelvic area. He did not report recent trauma or infection. The patient had no complaint about abdominal pain or diarrhea. His lesions had been previously treated using imiquimod and cryotherapy without noticeable improvement. The most severe consequence of his condition was the psychological burden on his sex life. No compression of the surrounding lymph vessels was seen at sonography of the pelvic area.

A biopsy was performed and showed dilated vascular channels lined by flat, thin cuboidal endothelium in the underlying superficial dermis. Acquired LC of the penis was diagnosed.

One session of electrocoagulation was performed under local anesthesia. This treatment allowed an almost complete regression of the lesion without recurrence after 3 years of follow-up (Fig. 2).

Discussion/Conclusion

LC is a vascular malformation resulting from a developmental anomaly of the superficial lymphatic system of the skin. It is uncommon and benign. LC generally occurs on the trunk, axilla, thigh, buttock area, and oral cavity, but less frequently on the penis [1]. LC may be either congenital or acquired. Acquired cases appear to develop most frequently after infections of lymphatic channels, irradiation, or surgery [1]. In a recent review of the literature, Macki et al. [2] reported only 8 cases of LC between 1947 and April 2018. Five of these were congenital and 3 were related to lichen planus, recurrent cellulitis, and ulcerative colitis. Since then, 3 others cases were reported: one case acquired post-radiotherapy for penile cancer and two others without history of trauma or surgery, irradiation, or previous infection [3, 4]. In our case, it was no previous surgery, radiotherapy, or underlying pathology.

At clinical examination, one may observe coalescent, translucent vesicles. Mild edema may be associated to the vesicles. The lesions are constantly painless.

LC is often mistaken for genital warts or molluscum contagiosum. In our case, the patient was misdiagnosed with genital warts and treated with cryotherapy and imiquimod without efficacy.
Due to the typical aspect of the lesions, a clinical diagnosis can easily be established, all the more when the clinician is aware of the disease [5]. However, a biopsy is often done to confirm the diagnosis and to choose suitable treatment.

At histological examination, superficial dilated lymphatic vessels located in the papillary dermis and mononuclear inflammatory cells throughout the papillary and reticular dermis [3] may be observed. In our case, both histological features were present.

Various treatments are available for LC including surgical excision (which is the gold standard), CO₂ laser ablation, cryotherapy, flash lamp pulsed dye laser, and electrocoagulation therapy [4]. Cryotherapy was not successful for the lesion of our patient. We thus decided to treat the lesions with electrocoagulation. We performed only one session under local anesthesia. It resulted in an almost complete recession of all the lesions. Toxicity included mild acute edema early after electrocoagulation. There was no sign of local recurrence after 3 years of follow-up.

Albeit asymptomatic, LC lesions on the penis may negatively affect the sexual life of the patients. Moreover, delayed diagnosis may be a source of psychological difficulties for them.

LC is a benign lesion with typical clinical presentation, whether it is congenital or acquired. However, penile location is rare. Electrocoagulation can be an effective treatment for these lesions. Electrocoagulation may be an efficient treatment for LC in locations that may be surgically challenging such as penis.

**Statement of Ethics**

The authors have no ethical conflicts to disclose. Informed consent was obtained from the patient. In accordance with the World Medical Association Declaration of Helsinki, the subject has given his written informed consent to publish his case.

**Disclosure Statement**

The authors have no conflicts of interest to declare.

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Fig. 1. Vesicles of the penile shaft and glans.

Fig. 2. Regression of the lesions after one session of electrocoagulation.