CASE REPORT
PEDIATRIC UROLOGY

Doi: 10.4274/jus.326
Journal of Urological Surgery, 2015; 3: 153-155

A Rare Case of Penile Lymphangioma Circumscriptum Treated with the Split-Thickness Skin Grafting Technique

Split-Thickness Dermal Greft Tekniği ile Tedavi Edilen Nadir Penil Lenfanjioma Sirkumskriptum Olgusu

Nurullah Hamidi, Onur Telli, Berk Burgu

Ankara University Faculty of Medicine, Department of Pediatric Urology, Ankara, Turkey

ABSTRACT

Lymphangioma circumscriptum (LC) is an uncommon type of microcystic lymphatic malformation involving the skin and mucosa caused by saccular dilatation of lymphatic channels. Patients may suffer from edema, vesiculopustular lesions and lymphatic leakage. There are many treatment methods for the management of lymphangioma circumscriptum. In this paper, we present a 10-year-old patient who had penile edema treated with split-thickness skin grafting technique.

Key Words

Penile edema, lymphangioma circumscriptum, split-thickness skin graft

ÖZET

Lenfanjioma sirkumskriptum (LS) deri ve mukozadaki lenfatik kanalların sakküler dilatasyonuna neden olan mikrokistik lenfatik malformasyonun nadir görülen çeşididir. Hastalarda ödem, vezikülopüstüler lezyonlar ve lenfatik akıntı görülebilir. Lenfanjioma sirkumskriptum tedavisinde bir çok yöntem mevcuttur. Bu olguda, Split-thickness dermal greft ile tedavi edilen 10 yaşındaki penil ödemi olan genç hastayı sunduk.

Anahtar Kelimeler

Penil ödem, lenfanjioma sirkumskriptum, split-thickness dermal greft

Introduction

Lymphangioma circumscriptum (LC) is a rare benign, hamartomatous developmental anomaly of the lymphatic vascular system that presents as translucent vesicles of varying size with a pink, red, or black hue. LC causes cosmetic problems besides refractory rupture, infection, lymphorrhea, and bleeding (1). These lesions may occur on any part of the body, but the axillary folds, shoulders, neck, proximal limbs, and buccal mucosa are the most common sites. LC is an unusual entity in the anogenital region and may be clinically indistinguishable from genital warts; consequently, biopsy from such lesions may be necessary to both confirm the diagnosis and formulate an appropriate treatment plan. Additionally, these lesions must be treated properly because they may act as a portal of entry for infections, be cosmetically embarrassing, and, if large, cause functional impairment (1,2).

Case Presentation

A 10-year-old boy was admitted with a history of penil edema (Figure 1a). The patient had a history of circumcision and bilateral orchiopexy 2 years ago. Edema was recognized at the 6th month of surgery and increased by time. Penile biopsy was performed and the lesions were histopathologically confirmed to be LC.

On examination, the penis shaft was covered with lymphedema and there were clusters of gelatinous-appearing, coalescent, verrucous vesicles and papules. There was no family history of any other lymphangioma. Under general anesthesia, a 8-F Foley urinary catheter was inserted to avoid injury to the urethra during the operation; circumcision incision was made and then, the affected area of the penis skin was excised. The wound bed was debrided to reduce bacterial contamination. Hemorrhage control was made for preventing graft rejection related to hematoma. Two pieces of skin taken from the bilateral inguinal region, with split-thickness skin graft (STSG). One of the grafts laid over the dorsal part and the other laid over the ventral part of the penis to leave the suture lines at the lateral sides of the penis (Figure 1b). In this way, a dorsal line on the penis was precluded and cosmetic problems were prevented. To provide immobilization, compression dressing was wrapped around the penis. At the 12th day, the dressing was completely opened and no hematoma or necrosis was observed. He was discharged at 12th day. At the 6th and 12th months of follow-up, there was no postoperative

Correspondence
Nurullah Hamidi MD, Ankara University Faculty of Medicine, Department of Pediatric Urology, Ankara, Turkey
E-mail: dr.nhamidi86@gmail.com **Received:** 26.02.2015 **Accepted:** 13.04.2015

complications and abnormality on the penis skin (Figure 1c). After complete surgical excision and the primary reconstruction, the cosmetic result was satisfactory.

Discussion

Lymphangiomatous lesions consists of dilated, superficial, lymphatic channels, which may occur either following damage to previously normal lymphatics or as a result of a congenital abnormality of the deep dermal and subcutaneous lymphatics. However, the clinical and histological features of these two types do not differ in their etiology (3). LC is generally localized at the axillary folds, neck and the chest and, rarely, at the genital area. LC is a widespread problem in the adult female population, but rare in the childhood (2). Different treatments, such as surgical excision, electrocoagulation, lasers, topical imiquimod, radiofrequency ablation and sclerotherapy have been used in with varying success (4,5). Kokcam has suggested "watch and wait" policy, which is advisable if the condition is asymptomatic (6). The only radical cure is to remove the superficial component as well as the deeper lymphatic cisterns. This is achieved through surgical destruction or laser ablation of the lesions. Several therapeutic modalities, such as electrocautery, cryotherapy, sclerosants, incision and drainage, and carbon dioxide laser have been tried but, in most of them, there is a high incidence of recurrence. Lymphangiosarcoma is a rare occurrence in LC; it has been reported at the site of an irradiated LC. Squamous cell carcinoma developing in a long-standing lesion has also been reported (7,8).

Figure 1a. Preoperative dorsal view of penis

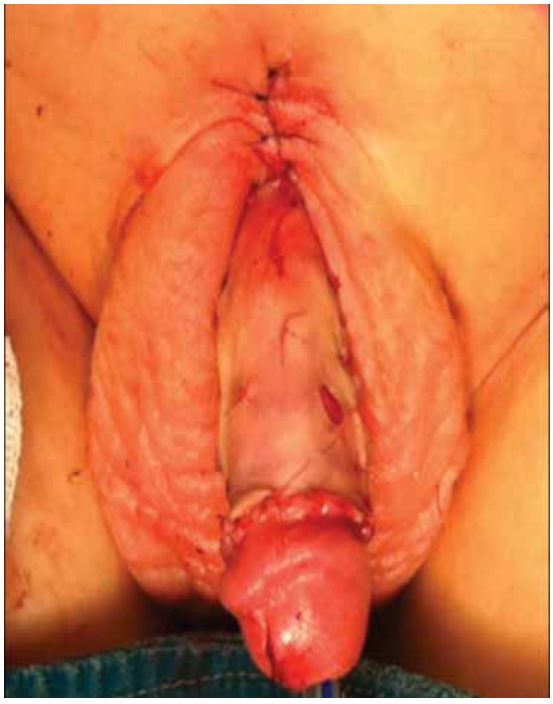

Figure 1b. Early postoperative dorsal view of penis after split-thickness dermal graft operation

STSGs are thinner and have less tissue, thus, it has less metabolic demand from the wound bed and has better survival response. In contrast, a full-thickness skin graft (FTSG) is thicker and it has greater metabolic demand. Survival of FTSG is more uncertain and it requires a well-vascularized recipient site. In addition, hair follicles are also present in the dermis. Therefore, since STSGs are generally hairless, they, are generally preferred on the penis (9). In the light of these facts, full-thickness excision of the skin and subcutaneous tissue down to the deep fascia still seems to be the best and most secure option. In this case, we tried to treat chronic penile lymphedema with STSG. We totally excised the penile skin with all subcutaneous tissue and replaced with STSGs instead of excised tissue. The patient had not any abnormality such as recurrence, fistula or necrosis during follow-up after surgery.

Conclusion

LC of the penis is an unusual entity that may be indistinguishable from genital warts. After confirmation of the diagnosis, a treatment

Figure 1c. 12th months after split-thickness dermal graft operation

plan consisting of wide excision should be outlined. STSG is the most appropriate choice for penile skin reconstruction because of the special characteristics of the penile skin, namely thin, hairless skin. Lesion excision and repair with STDG has been proved to be effective in terms of both function and cosmetic appearance in LC treatment. Using proper surgical technique and postoperative wound care maximizes graft success.

Informed Consent: Consent form was filled out by parents of patient, **Concept:** Onur Telli, Berk Burgu, **Design:** Nurullah Hamidi, **Data Collection or Processing:** Nurullah Hamidi, **Analysis or Interpretation:** Berk Burgu, **Literature Search:** Nurullah Hamidi, **Writing:** Nurullah Hamidi, Onur Telli, **Peer-review:** Internal peer-reviewed, **Conflict of Interest:** No conflict of interest was declared by the authors, **Financial Disclosure:** The authors declared that this study has received no financial support.

References

1. Yang X, Jin Y, Chen H, Li S, Ma G, Hu X, Qiu Y, Yu W, Chang L, Wang T, Lin X. Highly selective electrocoagulation therapy: an innovative treatment for lymphangioma circumscriptum. Dermatol Surg 2014;40:899-905.
2. Sasaki R, Negishi K, Akita H, Suzuki K, Matsunaga K. Successful treatment of congenital lymphangioma circumscriptum of the vulva with CO2 and long-pulsed Nd:YAG Lasers. Case Rep Dermatol 2014;6:1-4.
3. Latifoğlu O, Yavuzer R, Demir Y, Ayhan S, Yenidünya S, Atabay K. Surgical management of penoscrotal lymphangioma circumscriptum. Plast Reconstr Surg 1999;103:175-178.
4. Wang JY, Liu LF, Mao XH. Treatment of lymphangioma circumscriptum with topical imiquimod 5% cream. Dermatol Surg 2012;38:1566-1569.
5. Niti K, Manish P. Microcystic lymphatic malformation (lymphangioma circumscriptum) treated using a minimally invasive technique of radiofrequency ablation and sclerotherapy. Dermatol Surg 2010;36:1711-1717.
6. Kokcam I. Lymphangioma circumscriptum of the penis: a case report. Acta Dermatovenerol Alp Pannonica Adriat 2007;16:81-82.
7. Wilson GR, Cox NH, McLean NR, Scott D. Squamous cell carcinoma arising within congenital lymphangioma circumscriptum. Br J Dermatol 1993;129:337-339.
8. King DT, Duffy DM, Hirose FM, Gurevitch AW. Lymphangiosarcoma arising from lymphangioma circumscriptum. Arch Dermatol 1979;115:969-972.
9. Thakar HJ, Dugi DD 3rd. Skin grafting of the penis. Urol Clin North Am 2013;40:439-448.