



CASE REPORT

## A salvage surgical solution for recurrent lymphangioma of the prepuce

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### KEYWORDS

Lymphangioma; Trapped penis; Scrotum; Genital surgery

**Summary** Treatment of lymphangioma circumscriptum of the prepuce is often technically demanding. Many different medical and surgical approaches have been described over the years, none of which has proved completely successful. We describe a case in which skin deficiency was a problem after prior multiple excisions of lymphangiomatous tissue. We decided to bury the degloved shaft in the scrotum. After 6 months the penis was raised and reconstructed using scrotal skin; this achieved satisfactory skin coverage. In such cases piliferous follicles can easily be dealt with at a later stage by cosmetic treatment.

We believe this technical approach may be a valid alternative to conventional procedures (i.e. grafts and flaps) especially when extensive resection makes skin coverage particularly difficult.

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Lymphangioma circumscriptum (LC) is a rare primitive malformation of the lymphatic system that involves the skin and subcutaneous tissue. The commonest cutaneous form is characterised by multiple clusters of vesicles that contain clear lymph fluid. These clusters are likely to be superficial dilatations rising from lymph channels congenitally separated from the network of lymph vessels, for unknown reasons.<sup>1-3</sup> LC, usually noted within the first or second year of life, normally occurs on the proximal extremities, trunk, axilla, and oral cavity. In the genital region, and in particular the form involving the prepuce, LC has rarely been described. Not infrequently at this site,

the vesicles have a wart-like appearance and may be clinically indistinguishable from warts. Biopsy of such lesions may be necessary to confirm diagnosis and to formulate appropriate treatment.<sup>4,5</sup> Because of the high rate of recurrence, surgical excision has to be very extensive: therefore, significant problems can arise with reconstruction both from a functional and a cosmetic point of view.

The subject was a 16-year-old boy with a diffuse cystic coronal lesion of the residual prepuce; he had undergone three operations for LC, each followed by recurrence. His family history was unremarkable. Physical examination showed the coronal region to be covered with tense vesicles filled with clear fluid (Fig. 1).

Under general anaesthesia, lymphangiomatous

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**Figure 1** Penile appearance. The cluster of vesicles entirely surrounds the glans. Shaft skin is deficient with excess scar tissue.

and scarred tissue was excised. A tunnel was then created into the scrotum, where the denuded penis was buried (Fig. 2). After 6 months the shaft was raised and penile skin was reconstructed with the adjacent scrotal skin (Fig. 3). Fig. 4 shows the final result of the correction. The patient tolerated both procedures well, without postoperative complications. Histological examination confirmed the lymphangiomatous origin of the lesion. At 12-month follow-up, no negative psychological consequences were reported. The patient has successfully started cosmetic treatment for the local hairiness.

## Discussion

Despite its rare occurrence, lymphangioma circumscriptum has been repeatedly reported in the literature: it is more common in females and any cutaneous site may be affected, including the tongue.<sup>6</sup> Regarding the genital area, sporadic cases have been described involving the prepuce, glans and retropubic space.<sup>1,7,8</sup> The macroscopic appearance of LC is a cluster of a large number of vesicles of various size, containing lymph fluid.



**Figure 2** The penis is buried into the scrotal skin.

These vesicles represent superficial saccular dilations from underlying lymphatic vessels that occupy the papilla and push upwards against the overlying epidermis through vertical, dilated lymph channels, that are separated from the normal network of lymph vessels.

The reason for the failure of these primitive lymph sacs to connect to the rest of the lymphatic system is unknown.<sup>1,3</sup> Each skin lesion may range from a minute vesicle to a small bulla-sized lesion.



**Figure 3** Appearance immediately after reconstruction.



**Figure 4** At 3 months follow-up shaft skin is soft and elastic.

These lesions may appear at birth, but they are more often noted within the first to second year of life and usually occur on the proximal extremities, trunk, axilla and oral cavity.

Regardless of the anatomical site the vast majority of authors agree that the treatment of choice is early and adequate excision, since spontaneous involution is unlikely. Our case is consistent with this view since our patient had already experienced three failed attempts at excision: this was probably due to remnants of residual lymphatic tissue in the juxtacoronary internal layer of the prepuce, which had continued to grow. Our extensive resection of the lesion and scarred skin resulted in a completely degloved penile shaft: direct coverage with penile skin appeared to be impossible.

Many treatment options have been proposed over the years for reconstructing the genital region after surgical excision: in particular, reconstruction of the penis by skin graft requires multiple suture lines, particularly on the ventral side, and in a Z-like fashion, to avoid contraction;<sup>8</sup> on the other hand, tissue expanders are difficult to manage in the genital area. The use of flaps from the inguinal

region, from pubis, from the scrotum and the thigh, are more invasive, technically challenging and more prone to complications if compared to the two-stage approach using local scrotal skin. We believe that by using this procedure tissue transfer can be avoided thereby minimizing the risk of complications (i.e. suture ruptures, inadequate graft take).

The problem of scrotal hairy skin around the shaft can easily be dealt with after puberty by laser fulguration or electrocoagulation of piliferous follicles with little or no impact on cosmetic appearance.

In conclusion, the described two stage surgical correction of LC seems to be reliable and to be also suited to approaching other problems leading to severe trapped penis. It has proved to be effective in terms of both function and cosmetic appearance.

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