Annals of Case Reports

Massimo I, et al. Ann Case Rep: 7: 913. www.doi.org/10.29011/2574-7754.100913 www.gavinpublishers.com

Case Report

Surgical Treatment of Penile Lymphangiomatosis: A Case Report

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Citation: Iafrate M, Ruggera L, Sacchi D, Aceti A, Mancini M. (2022) Surgical Treatment of Penile Lymphangiomatosis: A Case Report. Ann Case Report 7: 913. DOI: 10.29011/2574-7754.100913

Received: 12 August 2022; Accepted: 16 August 2022; Published: 19 August 2022

Abstract

Lymphangiomas are uncommon hamartomatous malformations of the lymphatic system, that may affect several organs including bone, abdominal organs, lungs and skin. Skin lymphangiomas are most commonly found in the head and the neck, but the condition is rare in the genital organs. We report a rare case of multiple lymphangiomas of the penis, requiring highly specialized surgery in our Clinic [1].

Case Report

A 51-year-old man came to our attention with penile lesions at first diagnosed as fibromas. In history, the patient was in renal replacement therapy (3/week) and had undergone two renal transplants, in 1988 and 2002, due to glomerulonephritis; he also presented congenital muscular torticollis surgically treated during childhood.

Examination of the penis revealed several nodular lesions with two major groups, 3-4 cm each, in the ventral part of the penile shaft (Figures 1 and 2). It was decided to remove the two larger formations. Two skin lozenges were excised from the penis and, subsequently, a "Z" suture was applied to grant the continuity of the skin. No skin flap was necessary in order to cover the areas of defect. After surgery, the patient underwent a regular follow-up and after one month, the healing of the wound was complete (Figure 3 and 4).

The pathology of the surgical sample documented penile lymphangiomatosis, characterized by vascular malformation composed of large and dilated lymphatic channels separated by layers of connective tissue in the reticular dermis (Figure 5). The endothelial cells lining the surface of the cystic lymphatic channels were CD31 positive on immunohistochemistry. (Figure

6) At 2-years follow-up, the patient presented no recurrence of the lesions.

Discussion

Lymphangiomas are rare tumours of the lymphatic vessels which can affect different sites, such as abdominal organs, bones, skin (most commonly axilla, neck and proximal extremities), with the exception of the nervous system, which lacks in lymphatics [2-4].

Genital localizations of lymphangiomas are rare conditions, and may be difficult to diagnose and differentiate from genital warts or other infectious diseases [5-7]. Lymphangiomas usually present as asymptomatic, fluid-fill translucent lesions or vesicles; albeit not aggressive, they can seriously affect the patients' quality of life, especially causing pain, erectile dysfunction and psychological distress [8].

Incidence of lymphatic malformations is higher during infancy, with few cases occurring later in life. They may be congenital or acquired: the first are due to malformations of lymphatic vessels and mainly occur in childhood, usually before 5 years of age; acquired lesions are the results of local lymphatic damage after radiation, infections or pelvic surgery [3,9,10].

Volume 7; Issue 04

Ann Case Rep, an open access journal

ISSN: 2574-7754

Lymphangiomas may also be classified as superficial (lymphangioma circumscriptum) or deep (lymphangioma cavernosum and cystic hygroma).

Various treatment options for superficial lesions have been proposed, such as superficial radiation therapy, carbon dioxide laser, suction-assisted lipectomy, but all have shown a high rate of recurrence; some authors suggest not the treat superficial lesions as they may spontaneously resolve within few weeks [11]. For deeper lesions, surgery is the most effective treatment. It is important to radically remove lymphangiomas in order to reduce the recurrence rate, as high as 25% after the first excision. The main complications of surgical treatment are pain, and the formation of keloids [8].

There are just a few reported cases of lymphangiomas of the penis; treatment of such rare and complex conditions should be restricted to centres of experience, especially if connected in international networks of experts, such as the ERN eUROGEN, which includes our Hospital, where expertise can be shared and discussed in secure web-based platforms. In such centres, surgery is an effective and definitive treatment for penile lymphangiomas [1-12].



Figure 1: Several nodular lesions and two major groups, 3-4 cm each, in the ventral part.



Figure 2: Several nodular lesions and two major groups, 3-4 cm each, in the ventral part.



Figure 3: A "Z" suture was applied to grant the continuity of the skin.



Figure 4: A regular one-month follow-up and healing of the areas.

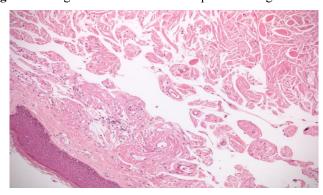


Figure 5: Thin and thick connective tissue in the reticular dermis.

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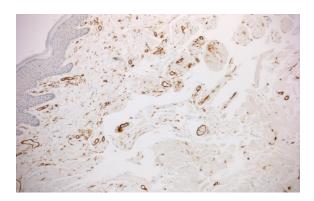


Figure 6: CD31 immunohistochemistry.

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