



Penoscrotal lymphangioma circumscriptum following circumcision

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ABSTRACT

We present a 15 year old boy with multiple vesicular lesions involving the scrotum and penile skin which developed following circumcision. Surgical excision of the penoscrotal skin with local flap cover was done. Histopathology was consistent with lymphangioma circumscriptum. To our knowledge this was the first case of penoscrotal lymphangioma circumscriptum which was seen after circumcision.

Keywords: Lymphangioma circumscriptum; penis; scrotum; surgery.

Introduction

Lymphangioma circumscriptum (LC) is a hamartomatous lymphatic malformation of deep dermal and subcutaneous lymphatic channels which can be congenital or acquired. It commonly presents in infancy but can occur at any age. Patients present with multiple vesicular lesions which are pink to copper coloured.^[1] Treatment is for cosmetic reasons and for risk of recurrent episodes of cellulitis. Surgical excision is the treatment of choice. After surgery patients need close follow up since there is a high risk of recurrence. Lymphangioma circumscriptum of penoscrotal region is rare.^[2] We present a 15 years old boy with penoscrotal lymphangioma circumscriptum developed post circumcision which was managed with surgical excision and local flap cover.

Case presentation

A 15 years old boy presented to the urology outpatient department with history of multiple fluid filled lesions involving the scrotum and penile skin since 8 months. These lesions were progressively increasing in number and would discharge serous fluid after spontaneous rupture. He also gave history of recurrent episodes of mild fever associated with pain and redness in the involved area. Detailed history revealed

that patient underwent circumcision one year back and the lesions developed after that.

On local examination there were multiple translucent vesicles over scrotum extending onto the penile shaft. Underlying scrotal and penile skin was diffusely thickened (Figure 1). On rupturing these vesicles there was oozing of serous fluid. There was no associated inguinal lymphadenopathy or lymphedema of legs. Routine haemogram, liver function tests and renal function tests were within normal limits. Filariasis serology was negative. Scrotal ultrasonography showed normal bilateral testes and cord structures. Wedge biopsy of the scrotal skin taken and sent for histopathological examination. Histopathology revealed presence of multiple dilated endothelial lined lymphatics (Figure 2). Thus a diagnosis of lymphangioma circumscriptum was finally made.

Patient was managed with complete excision of the skin and subcutaneous tissue of penis and scrotum (Figures 3 and 4). The resulting defect with the exposed testes was closed by raising flap from the inner thighs bilaterally and closing them in the midline. Penile shaft was covered with split thickness skin graft harvested from left thigh (Figure 5). His postoperative course was uneventful and he is being followed up regularly for any recurrences.

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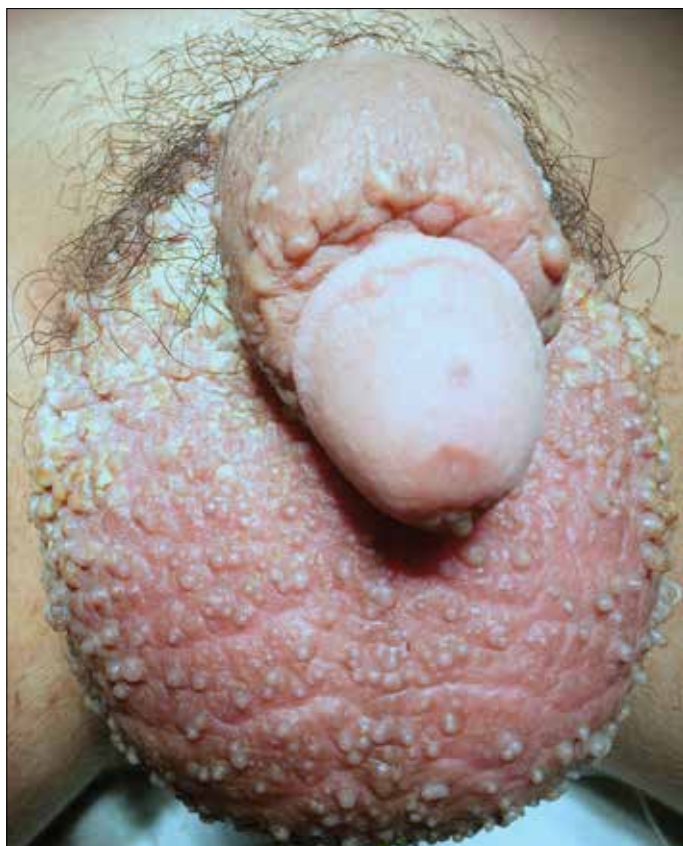


Figure 1. Multiple vesicular lesions involving the entire scrotum and penile skin

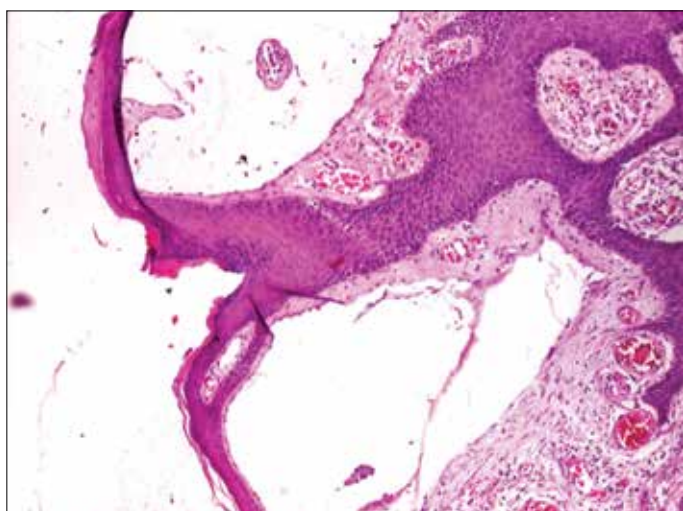


Figure 2. Histopathology showing multiple dilated lymphatic channels

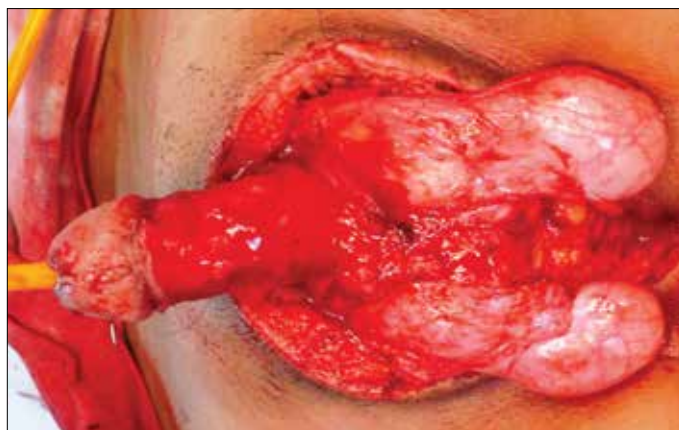


Figure 3. Intraoperative photograph after excision of involved penoscrotal skin



Figure 4. Excised penoscrotal skin intoto after surgery



Figure 5. Reconstructed penile and scrotal wound at the end of the surgery

Discussion

Lymphangiomas are uncommon, congenital, hamartomatous malformations of lymphatic system which involve skin and subcutaneous tissue. On the basis of depth and size of abnormal lymph vessels lymphangiomas can be either superficial such as lymphangioma circumscriptum or deep which include cavernous lymphangioma and cystic hygroma.

Lymphangioma circumscriptum is characterized by presence of multiple clusters of translucent vesicles containing clear lymph fluid often referred to as frog spawn. Sometimes due to secondary haemorrhage vesicles appear pink or red. These vesicles represent saccular dilatations from underlying lymphatics which are connected to deep feeding cisterns. These cisterns have a thick coat of muscle fibres which rhythmically contract and thus cause dilatation of superficial lymphatic channels by increasing the intramural pressure. This pathogenesis was proposed by Whimster in 1976.^[1]

Common sites of predilection are extremities, axilla, trunk and oral cavity.^[3,4] Involvement of scrotum though reported is rare. Congenital LC is commonly seen in infancy but can occur at any age. Acquired LC is seen in elderly following radiotherapy, surgery like vasectomy and infections like filariasis, tuberculosis and lymphogranuloma venereum.^[5-9] Our patient presented at the age of 16 years and had history of undergoing circumcision prior to onset of lesions. Manhandling of tissue during circumcision could have led to ligation of draining lymphatics. Though the exact pathogenesis remains obscure.

Patients can be asymptomatic or can present with history of oozing of serosanguinous fluid. At times recurrent episodes of cellulitis characterized by fever and pain in affected area can complicate the disease course. Similar history of oozing of fluid and cellulitis was present in our patient. Common conditions which pose diagnostic difficulty include molluscum contagiosum and warts necessitating histopathological confirmation. Skin biopsy done in the presented case helped in clinching the final diagnosis. Further, it helped in deciding the treatment plan. Management includes laser ablation using argon laser, carbon dioxide laser, diode laser or pulsed dye laser. Further sclerotherapy and radiotherapy can also be tried. In asymptomatic patients close observation is kept.^[10-12] Indications for surgery include recurrent episodes of cellulitis, persistent discharge from ruptured vesicles or for cosmetic concerns. Surgical excision gives the best chance of cure. It involves excision of skin and subcutaneous tissue till the deep fascia. Inadequate excision leads to recurrence of lesions. Complete surgical excision upto deep fascia was done in our patient and the defect was closed by raising flaps from inner thighs bilaterally. Post-operative result was cosmetically acceptable.

This case is being presented to highlight the importance of keeping LC as differential diagnosis in case of patients presenting with vesicles over scrotum especially after some surgical intervention. Further the need for histopathological confirmation is stressed upon. Surgical excision should be complete so as to remove all the feeding cisterns to decrease the chances of recurrence.

Informed Consent: Written informed consent was obtained from patients' parents and patient who participated in this case.

Peer-review: Externally peer-reviewed.

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