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MANAGEMENT OF PEDIATRIC PENOSCROTAL LYMPHEDEMA WITH SCROTAL DEBULKING AND VASCULARIZED OMENTAL LYMPH-NODE TRANSFER: A CASE REPORT

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SUMMARY

Penoscrotal lymphedema is a rare but debilitating condition characterized by swelling of the penis and scrotum due to lymphatic fluid accumulation. This condition can lead to significant physical and psychosocial issues.

This report presents the case of an 11-year-old boy with penoscrotal lymphedema successfully treated with Ablative and physiological surgery. Treatment included scrotal debulking and Vascularized Omental Lymph-node Transfer (VOLT) based on the right gastroepiploic artery, anastomosed to the descending branch of the left lateral circumflex femoral artery.

The patient experienced an uneventful recovery with improvement of symptoms and quality of life by the 6th month. In conclusion, this case highlights the value of integrating ablative and physiological techniques, such as Vascularized Omental Lymph- node Transfer (VOLT), for the effective management of penoscrotal lymphedema, particularly in patients necessitating lymph node transfer to multiple recipient sites.

Keywords: Penoscrotal lymphedema, Debulking, Vascularized omental lymph-node transfer, Case report.

INTRODUCTION

Lymphedema is characterized by persistent tissue swelling resulting from impaired lymphatic drainage (1). It can affect any part of the body including the genitals. Penoscrotal lymphedema occurs when the swelling affects both the scrotum and the penis. Lymphedema may occur as a primary condition i.e, due to congenital abnormalities in lymphatic development; or secondarily e.g., following trauma, infection, or surgical disruption of lymphatic channels(1)(2). The resultant accumulation of lymphatic fluid triggers a chronic inflammatory response, with subsequent deposition of adipose

and fibrosis, contributing to tissue enlargement over time(1)(2) (3). Opinion regarding management is contentious, with a split between a conservative approach and surgical route(2,4).

The optimal timing for surgical intervention is controversial. Some have opted for early treatment strategies aimed at minimizing the physical and psychosocial impact on affected children (5), while others have preferred a delayed approach waiting until pubertal changes to avoid overcorrection as well as to tap onto the benefit of operating on larger structures. A combined approach is also possible with an early debulking followed by additional physiological procedures when the child is older (3).

CASE PRESENTATION

11-year-old male with a four-year history of scrotal and penile swelling. Ultrasound revealed penile edema, chronic infection, and lymphoscintigraphy confirmed primary lymphedema. Despite conservative management with compression therapy and oral/topical antibiotics the condition progressed, leading to massive scrotal swelling which extended to upper thighs with skin changes, causing discomfort and major psychosocial effects. No history of trauma, surgery, recurrent infections, malignancy or family history of similar conditions. On examination, there was notable non pitting scrotal swelling involving the penis and bilateral thighs proximally and thickened hyperpigmented scrotal skin with some lymph-angiomatous lesions as demonstrated by figure1.

Figure 1; pre-operative



Figure 2: Omental flap, (hemostat showing right gastroepiploic artery)

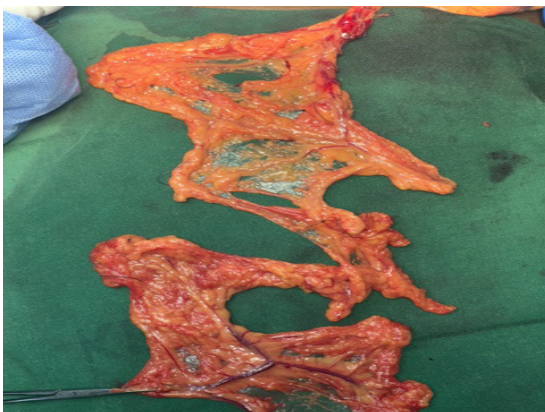
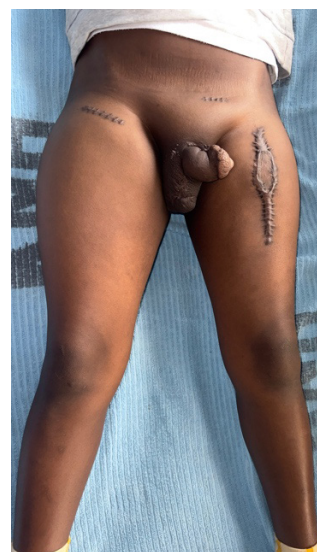


Figure 3; immediate post-operative



Figure 4; 6-months post-operation



Both surgical ablative and physiological techniques were done. Scrotal debulking using the Homan's procedure and VOLT based off the right gastroepiploic vessels which was anastomosed to the descending branch of the left lateral circumflex femoral artery. The flap was strategically distributed across the scrotum and bilateral thighs. The wound over anastomosis was partly left open, to avoid pressure which was later grafted with split thickness graft.

RESULTS

The patient was evaluated at 2 months and 6 months postoperatively. Table 1&2 present assessments of the dimensions and changes in quality of life using the Lymphedema Quality of life (LYMQOL) instrument adapted for genital lymphedema.

Table 1: Results

	Pre-operation (cm)	2nd-months post operation (cm)	6-months post operation (cm)
Right thigh (18 cm from the ASIS)-circumference	52,5	50.5	44
Left thigh-circumference	53	52	42
Scrotal-length	41	12	12
Scrotal-circumference	39	18	10
Penile-length	11	11	11
Penile mid-shaft circumference	11	10	9

Table 2: Preoperative and Postoperative LYMQOL Scores

Domain	pre-op score	6th -months post-op score
Function/4	3	1
Appearance/4	4	2
Symptoms/4	4	1
Mood/4	4	1
Overall QoL/10	2	9

At six-months after surgery, the patient showed significant improvement in all LYMQOL quality of life domains. The functional score decreased from 3 to 1, as the patient could perform daily activities and play comfortably with his peers. Appearance improved from 4 to 2 with reduced scrotal and thigh size and better skin pliability. Symptoms such as scrotal heaviness, skin irritation, and recurrent infections were eliminated, lowering the score from 4 to 1; similarly, mood scores improved. Overall, quality of life was greatly improved from 2 to 9.

DISCUSSIONS

Pediatrics genital lymphedema is uncommon and pose significant physical and psychological burdens, frequently presenting with penoscrotal involvement, as was the current case (3). The scrotum is affected in most cases, whereas isolated swelling of the penis alone is uncommon(3). Early disease presents with pitting edema. Progression leads to fibrosis complicating management(3).

Management of lymphedema depends on the severity of the condition and availability of resources. Management can either be conservative or surgical. The primary goal of conservative treatment is to minimize stagnant lymph accumulation to prevent

progression. It offer temporal relief but inadequate in advanced stages(3). For patients with lower limb lymphedema, compression stockings are the standard approach in non- operative care but with genital lymphedema because of the shape and location, stocking material is difficult to apply(5,6).

Surgery is indicated in cases of failed conservative management, disfigurement and functional impairment(3). It remains the definitive management for advanced-stage (6). There are two main surgical options: Ablative or physiological.

Ablative procedures remove lymph- edematous tissue with or without excision of the overlying skin. Ablative surgeries includes liposuction and surgical excision (6). In cases where tissues are fibrotic, liposuction might not be effective, as demonstrated in this case where the scrotal tissue was already fibrosed. Besides decreasing tissue volume, one benefit of ablative surgery is its ability to improve appearance(6). However, it is important to note that this approach does not address the root cause of the problem.

Physiologic procedures focus on restoring lymphatic function , Vascularized lymph node transfer (VLT) is one of the physiological procedure (7,8). Various mechanisms have been proposed to explain how vascularized lymph node transfer (VLNT) works. One such mechanism is the pumping effect, which is driven by the perfusion gradients between interstitial pressure and venous outflow.(9). Transplanted lymph nodes also secrete growth factors like vascular endothelial growth factors-C (VEGF-C) which induces local lymph angiogenesis(7).

In this case VOLT was used. VOLT offers a low donor site morbidity and negligible donor site lymphedema unlike traditional VLNT sites such as the groin, axilla regions (7). As noted by Rachel *et al.*, 2023, VOLT has consistent size of the gastroepiploic vessels running longitudinally through the flap. This feature allows the omentum to be divided into several flaps. So

far, the omentum is the only donor site capable of being transferred to multiple recipient areas(7)(6), as demonstrated by the case in which a long omental flap (figure 2) was harvested and distributed to bilateral thighs and the scrotum with a single anastomosis site. The omentum also contains abundant lymph nodes and lymphoid-reticular structures called 'milky spots along with its immunogenic and angiogenic properties which enhance lymphatic drainage(6,7,9).

CONCLUSION

This case highlights the effectiveness of VOLT and debulking surgery in managing extensive pediatrics penoscrotal lymphedema, especially where multiple recipient sites are involved. It also demonstrates the benefit of early surgical intervention in cases where conservative measures have proven inadequate, as it significantly improves the patient's physical function and quality of life as demonstrated by above results.

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