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Rare case of giant lymphocele treated with supramicrosurgical approach

INTRODUCTION: *Lymphocele (or cystic lymphangioma) is a typical disease of the lymphatic vascukarization caused by lymphatic fluid leakage. Lymphatic leakage can result from traumas or as a complication of surgical procedures. Clinic is vague and surgical resection is still considered the most effective approach. Thereby, a standpoint should be the identification and treatment of afferent lymphatic channels which can be addressed by LVA.*
CASE PRESENTATION: *The authors describe a rare case of a giant lymphocele occurred in a 56-year-old Caucasian woman treated for endometrial carcinoma. Lymphocele was refractory to percutaneous drainage and compressive treatment. Therefore surgical excision in combination with supramicrosurgical lymphatico-venular anastomosis (LVA) was scheduled.*
CONCLUSIONS: *The aim of the report is to offer an overview on the main therapeutic options to treat lymphocele and to demonstrate the effectiveness of combining excision with lymphatic microsurgery.*

KEY WORDS: Inguinal lymphocele, LVA, Supramicrosurgery

Introduction

Lymphocele is a pathological condition caused by the extravasation of lymphatic fluid from lymphatic vessels. The symptomatology is vague (oppression and pain) and often leads to a delayed identification of the disease.
Main risk factors include lack of ligation of lymphatic vessels, radiation therapy, metastasis to the LNs and heparin therapy .
Ultrasonographic examination represents the primary step for the diagnosis of lymphocele. Likewise, CT scan and MRI appears to be feasible technique not only for diagnosis but also for the evaluation of lymphatic network.

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Case Report

The authors report a case of a recurring inguinal lymphocele following hysterectomy and inguino-femoral lymphadenectomy. for which dye mapping of lymphatic leakage and subsequent surgical groin exploration and ligation of the vessels was scheduled.
A 56-year-old woman underwent modified radical hysterectomy with bilateral inguinofemoral lymph node dissection for endometrial carcinoma.
Two months after dismission, the patient complained of swelling and tenderness in the right groin region. CT scan was taken and lymphocele diagnosis was made (Figs. 1 A, B, C)
Surgical excision was then scheduled in combination with supramicrosurgical LVA in order not only to address the afferent lymphatic vessels but also to reduce lymphatic pressure in the limb. Intraoperative ICG lymphography was performed (Fig. 2). Multiple 0,1 ml injection of ICG dye were performed in the foot and around the groin area. Lymphatic drainage was studied using an

ABBREVIATION

LVA: lymphatic venous anastomosis
LNs: lymph nodes
MR: magnetic resonance
CT: computed scan
ICG: indocyanine green

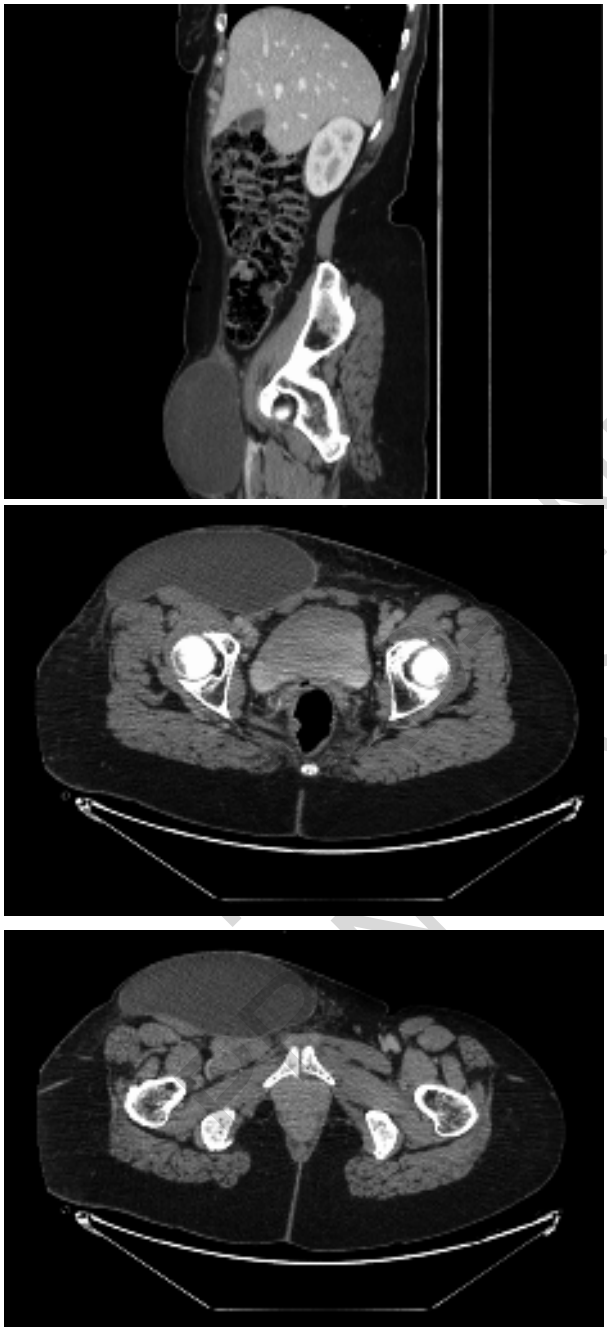

Fig. 1: A,B,C) Preoperative CT scan demonstates the presence of lymphocele located in the inguinal region.

Fig. 2: Preoperative ICG lymphography demonstrates dermal backflow in the distal portion of the limb resulting from lymphatic compression caused by the lymphocele.

Fig. 3: Intraoperative picture of the excised lymphocele. Its dimension were 20x9 cm.

infrared camera system (Photodynamic Eye (PDE); Hamamatsu Photonics, Japan).
Through meticulous microsurgical dissection, the main vascular and nervous structures of Scarpa triangle were identified and preserved. Lymphocele laid just over the femoral nerve, femoral vein and artery on which it was attached. Complete excision of the lymphocele with its capsule was therefore obtained. Dimension of lymphocele were 20 x 9 cm. (Figs. 3, 4).
Subsequently, supramicrosurgical LVA were performed. Creation of lymphaticovenular bypasses were perfomed not only addressing lymphatic vessels supplying lymphocele and causing accumulation of fluid but also distally in the lower portion of the limb in order to reduce the lymphatic system pressure locally and avoid any further risk of developing secondary lymphedema (Fig. 5).

Fig. 4: Intraoperative: lymphocele adhered to femoral blood vessels

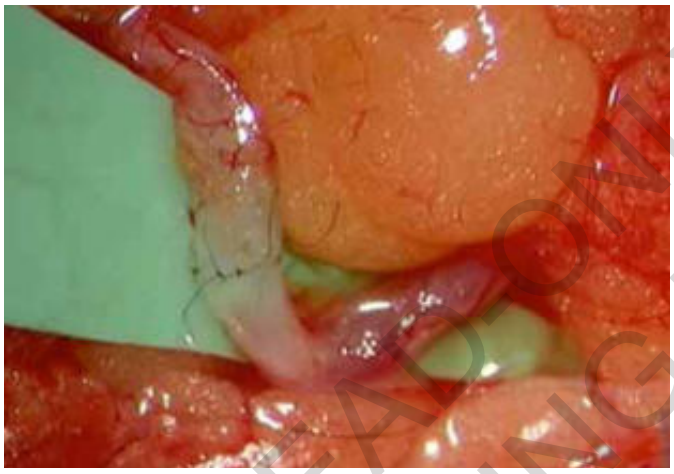

Fig. 5: Lymphatic vessel was identified distally from lymphocele and lymphatico-venular anastomoses were completed.

The patient had drainage for six days and compressive medication. No recurrence occurred.

Discussion

Lymphocele is a pathological condition caused by leakage of lymphatic fluid [1].
Inguinal lymphocele represents the most frequent postoperative complication of pelvic lymphadenectomy, which is a crucial step in gynecological cancer [2].
Formation of inguinal lymphocele after histerectomy is a common occurrence: incidence stands at 10-25%.
Lymphatic fluid may accumulate in either or both sides of the urinary bladder, filling the spaces in the lateral pelvic wall. Accumulation of large volume up to 1 litre can occur [2]. Risk factors include: extensive pelvic lymphadenectomy, lack of lymphatic vessel ligation, preoperative or postoperative radiation therapy, presence of metastasis to the LNs, use of retroperitoneal suction drainage, and administration of low-dose heparin for thromboembolic prophylaxis [3,4].
The non-specificity of symptomatology represents a contributing factor in delayed diagnosis; and, since lymphoceles are molstly incidental findings, there is no standard diagnostic protocol.
Oppression and pain may occur due to the growth of the neoformation.
Compression of neighboring pelvic organs, obstructive disease of urinary district, cystitis and complications occur mostly later, due to its large cavity size [5].
Differential diagnosis can include: urinoma, hematoma, cystic lymphangioma, neoplasm.
According to guidelines, ultrasonography is the primary step for the diagnosis of lymphoceles: it appears as an anechoic cystic structure and it may contain thin septations and debris. Pelvic lymphoceles can be wide and extend to retroperitoneum. Lymphoceles are seen as thin-walled hypodense lesions with water attenuation valued on CT imaging usually adjacent to surgical clips in patients who underwent lymphadenectomy. They are typically well defined, round or oval and uniformly dense. Infected or complicated lymphoceles usually have a thick irregular enhancing wall. On MR imaging lymphoceles appear as lobulated highly hyperintense structures with imperceptible wall and negligible wall enhancement [6].
ICG lymphography can detect lymphatic leakage, lymphatic hypertension and lymphatic dermal backflow.
To date, there are various treatment options for post-operative lymphoceles, including percutaneous drainage, sclerotherapy, percutaneous image-guided lymphatic ligation and fibrin glue injection. According to literature, percutaneous method in conjunction with sclerotherapy can be considered as first line treatment modality for lymphoceles due to its effectiveness, easeness of procedure and low complication rate. Drainage can relieve symptoms by decreasing the accumulation of lymphatic fluid.
Among the pharmacological options, somatostatin and its analog octreotide are highly effective in reducing lymphatic production and decreasing lymphatic flow, although the mechanism is still not clear; on the other hand the most effective sclerotizing agents and percutaneous glue are: doxycicline, tetracycline, bleomycin and povidone-iodine [7].
Excision surgery with complete removal of the lymphocele appears to be the elective treatment. Furthermore supramicrosurgical LVA can be combined with excision surgery to reduce the risk of recurrence [8]. In fact, recurrence occurs if lymphatic leakage continues, so that lymphaticovenular anastomoses can be adopted to address the leakage. Many techniques for treating lymphatic pathologies have been proposed; among them, recently, enteromesenteric bridge with the omental transposition [9].

LVA has been widely reported to be an effective procedure for treating lymphedema, lymphangitis and other lymphatic pathologies [10-16].

Conclusions

Inguinal lymphocele appears to be a common complication following major abdominal surgery. It should be adequately treated since its high recurrence rate. Surgical excision combined with lymphatico-venular anastomosis represent and effective therapeutic option with low risk of potential recurrence.

Riassunto

Il linfocele (o linfangioma cistico) è una malattia tipica della vascolarizzazione linfatica causata dalla fuoriuscita di liquido linfatico. La perdita di linfa può derivare da traumi o come complicanza delle procedure chirurgiche. La clinica è vaga e la resezione chirurgica è ancora considerata l'approccio più efficace. Pertanto, un punto importante dovrebbe essere l'identificazione e il trattamento dei canali linfatici afferenti che possono essere affrontati con un'anastomosi linfo-venosa.
Gli autori descrivono un raro caso di linfocele gigante verificatosi in una donna caucasica di 56 anni trattata per carcinoma endometriale. Il linfocele era risultato refrattario al drenaggio percutaneo e al trattamento compressivo. Pertanto è stata programmata l'escissione chirurgica in combinazione con anastomosi linfatico-venulare ultra microsurgica (LVA).
In conclusione si descrive una panoramica delle principali opzioni terapeutiche per il trattamento del linfocele e la dimostrazione dell'efficacia della combinazione dell'escissione seguita da ricostruzione con microchirurgia linfatica.

References

1. Hazir B, Haberal HB, Akinci D, Akdogan B: *An unusual localization of seven months delayed pelvic lymphocele following radical retropubic prostatectomy. Case report and literature review.* Int J Surg Case Rep, 2018; 44:181-84.

2. Kim HY, Kim JW, Kim SH, Kim YT, Kim JH: *An analysis of the risk factors and management of lymphocele after pelvic lymphadenectomy in patients with gynecologic malignancies.* Cancer Res Treat, 2004; 36(6):377-83.

3. Tsuda N, Ushijima K, Kawano K, Takemoto S,Nishio S, Sonoda G, Kamura T: *Prevention of lymphocele development in gynecologic cancers by the electrothermal bipolar vessel sealing device.* J Gynecol Oncol, 2014; 25(3):229-35.

4. Park SC, Lee JW, Park SA, Lee YO, So BJ, Rim JS: *The deep vein thrombosis caused by lymphocele after endoscopic extraperitoneal radical prostatectomy and pelvic lymph node dissection.* Can Urol Assoc J, 2011; 5(3):E40-E43.

5. Weinberger V, Fischerova D, Semeradova I, Slama J, Cibula D, Zikan M: *Ultrasound characteristics of a symptomatic and asymptomatic lymphocele after pelvic and/or paraaortic lymphadenectomy.* Taiwan J Obstet Gynecl, 2019; 58(2):266-27.

6. Lv S, Wang Q, Zhao W, Han L, Wang Qi, Batchu N, Ulain Q, Zou J, Sun C, Du J, Song Q, Li Q: *A review of the postoperative lymphatic leakage.* Oncotarget, 2017; 8(40): 69062-9075.

7. Yildirim IO, Berktas B, Saglik S, et al.: *Management of non-vascular complications following renal transplantation using percutaneous approach*. Ann Ital Chir, 2018; 89:86-91.

8. Mihara M, Hara H, Furniss D, Narushima M, Iida T, Kikuchi K, Ohtsu H, Gennaro P, Gabriele G, Murai N: *Lymphaticovenular anastomosis to prevent cellulitis associated with lymphoedema.* Br J Surg, 2014; 101(11):1391-96

9. Borz C, Muresan M, Jimborean O, et al.: *Modified enteromesenteric bridging operation for primary lymphedema.* Ann Ital Chir, 2018; 89:350-56.

10. Gennaro P, Gabriele G, Salini C,Chisci G, Cascino F, Xu FJ, Ungari C: *Our supramicrosurgical experience of lymphaticovenular anastomosis in lymphoedema patients to prevent cellulitis.* Eur Rev Med Pharmacol Sci, 2017; 21: 674-79.

11. Gennaro P, Borghini A, Chischi G, Mazzei FG, Weber E, Tedone Clemente E, Guerrini S, Gentili F, Gabriele G, Ungari C, Mazzei MA: *Could MRI visualize the invisible? An Italian single center study comparing magnetic resonance lymphography (MRL), super microsurgery and histology in the identification of lymphatic vessels.* Eur Rev Med Pharmacol Sci, 2017; 21(4):687-94.

12. Gennaro P, Gabriele G, Aboh IV, Cascino F, Zerini F, Aboud MG: *Ultramicrosurgery: A new approach to primary male genital lymphedema.* JPRAS Open, 2019; 20:72-80.

13. Gennaro P, Gabriele G, Mihara M, Kikuchi K, Salini C, Aboh IV, Cascino F, Chisci G, Ungari C: *Supramicrosurgical lymphaticovenular anastomosis (LVA) in treating lymphoedema: 36-months preliminary report.* Eur Rev Med Pharmacol Sci, 2016; 20: 4642-653.

14. Gabriele G, Aboh IV, Cascino F, Zerini F, Amadi JU, Del Frate R, Xu J, Gennaro P: *Indocyanine green sentinel node in merkel cell carcinoma of the cheek.* J Craniofac Surg, 2019; 30(4):e376-e377

15. Mihara M, Hara H, Kikuchi K, Yamamoto T, Iida T, Narushima M, Araki J, Murai N, Mitsui K, Gennaro P, Gabriele G, Koshima I: *Scarless lymphaticvenous anastomosis for latent and early-stage lymphoedema using indocyanine green lymphography and non-invasive instruments for visualising subcutaneous vein.* J Plast Reconstr Aesthet Surg, 2012; 65(11):1551-8

16. Barone V, Borghini A, Tedone Clemente E, Aglianò M, Gabriele G, Gennaro P, Weber E: *New insights into the pathophysiology of primary and secondary lymphedema: Histopathological studies on human lymphatic collecting vessels.* Lymphat Res Biol, 2020; Epub ahead of print.