

Modified enteromesenteric bridging operation for primary lymphedema



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BACKGROUND: Primary lymphedema represents a condition which affects the lymph vessels and their cells without any pathologic patient history leading to soft tissue swelling. They are described in the literature three types: congenital primary lymphedema, or Milroy disease, lymphedema praecox, which occurs in puberty, and lymphedema tarda, when, the same symptoms, develop over 35 years old patients. The aim of this paper is to present a modified Kimmoth's original procedure for a praecox lymphedema by introducing an omental flap in the resorptive area.

CASE REPORT: We present a case of 18 year-old patient with a significant lymphedema of the lower limbs and genitalia associated with high-symptomatic under-knee and scrotal ulcerative, necrotic and inflammatory lesions. The ultrasound and computed-tomography scans showed bilateral hypertrophic lymph-nodes in the inguinal and external iliac areas. Considering this diagnosis we decided to perform a physiologic lymph drainage by combining the enteromesenteric bridge with the omental transposition. The postoperative course was favorable without wound complications. Three months after, it was resected the remnant scrotal sclero-lymphatic tissue. 3 years after surgery, the clinical follow-up showed a significant reduced lymphedema with 10 cm shank / 15 cm thigh circumference lost.

CONCLUSIONS: The enteromesenteric bridge combined with the omental flap proved to be efficient in ensuring the lymphatic drainage in a case with proximal lymphatic occlusion.

KEY WORDS: Enteromesenteric bridging operation, Omental flap transposition, Primary lymphedema

Introduction

The enteromesenteric bridging operation (Kinmoth's operation) is known as a surgical option used for the treatment of the primary lymphedema ¹. The brilliant principle of this operation is to bypass a retroperitoneal lymphatic occlusion by using an alternative path: the functional mesenteric lymphatic circulation. In the medical literature, there are few reports concerning this issue²⁻⁵. By introducing an additional area for lymph resorption, we modified the enteromesenteric bridging

operation and applied it in a patient with "praecox" lymphedema. We present the main data which led us to the diagnosis of "praecox" primary lymphedema due to a proximal lymphatic occlusion in an 18 year-old male patient, and the modified enteromesenteric bridging operation performed. The patient's consent was obtained for publication of the report as well as for the procedure itself.

Method

CASE PRESENTATION

DIAGNOSIS

- **Clinical features:** the 18 years-old patient was admitted with a significant lymphedema of the lower limbs and genitalia associated with high-symptomatic under-

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Fig. 1

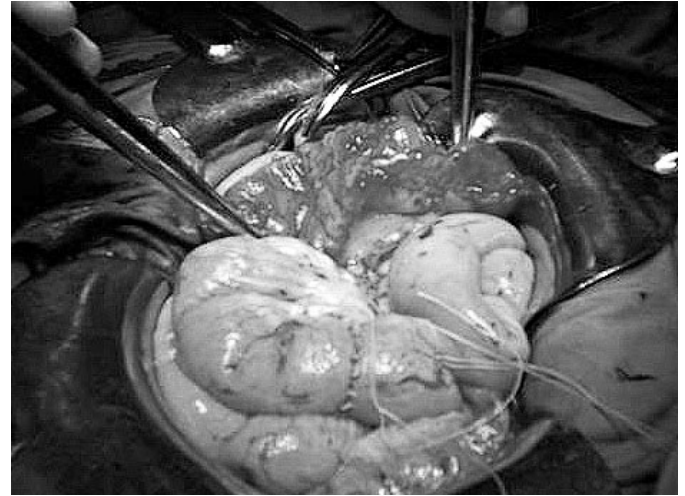


Fig. 2

knee and scrotal recurrent infections with ulcerative, necrotic and inflammatory lesions (Fig. 1). These significant symptoms appeared 2 months before the hospital admission and they determined the patient to ask medical advice.

The onset of the lymphedema was at the age of 4 years and was not preceded by any significant diseases or conditions as infections, tumors, trauma, previous surgical or radiological treatments. These clinical aspects allowed the diagnosis of “*praecox*” primary lymphedema². The involvement of both limbs and genitalia already sustains the diagnosis of proximal primary hypoplastic lymphedema.

- *Imaging features*: the ultrasound and computed-tomography scans showed bilateral hypertrophic lymph-nodes in the inguinal and external iliac areas and no lymphatic masses proximally, periaortic or around the common iliac vessels. A radionuclide lymphoscintigraphy was unavailable, but we considered that the computed-tomography scans associated with the clinical signs were eloquent for diagnosis of proximal lymphatic occlusion. There were no signs of mesenteric lymphatic occlusion: there were no signs of enteromesenteric lymphedema; furthermore, the patient didn't have any sign of malabsorption.

- *The diagnosis based on clinical and radiological features*: “*praecox*” primary lymphedema due to proximal lymphatic occlusion.

- *Treatment decision*: considering this diagnosis we decided to perform a physiologic lymph drainage, combining the enteromesenteric bridge with the omental transposition^{3,4}.

SURGICAL TREATMENT

The goal of our operation was to create lymphatic fistulae from the hypertrophic lymph nodes located in

external iliac artery areas and to drain these fistulae through an enteromesenteric bridge and through omental lymph vessels. This surgical strategy allows the lymphatic drainage from a wide lymphatic tributary territory including both inferior limbs and perineal organs. The original Kinmoth's enteromesenteric bridging operation would have ensured only an uni-lateral lymphatic drainage, for only one inferior limb and with poor results for the perineal edema.

In order to prevent post-surgery complications, such as a lymphocele, a potentially huge lymph pool or a pelvic abscess we associated prophylactic measures: antibiotics for minimum 7 days post-surgery and drain tubes in pelvic area until getting a significant decrease of the external drainage content. In addition, we introduced inside the lymphoreic area an omental flap, both for its absorptive and especially for its antiinfection properties.

DETAILED SURGICAL PROCEDURE

- *Isolation of the enteromesenteric bridge*: after a median celiotomy at inspection we did not notice any anatomic sign of mesenteric lymphatic stasis: the anatomic aspect of the mesentery and of the intestine was normal, non-edematous. This observation strengthened our decision to perform an enteromesenteric bridging operation.

We isolated a 15 cm-long segment of the mid-ileum together with its mesenterial arterial, venous and lymphatic vascularization. The small bowel continuity was reconstructed by a double-layer end-to-end anastomosis. The vascularized isolated intestinal segment was opened longitudinally and we removed the mucosal layer of the intestinal wall using a not too sharp curette: so there was exposed the submucosal layer which has an absorbance function due to its lymphatic vessels ends; so we obtained an 10x15 cm “absorptive” rectangle (Fig. 2).

- *Creation of a lymphoragic area in the pelvi-subperitoneal*

region. We removed a 20x20 cm rectangle of the median pelvic peritoneum. In this rectangular subperitoneal area we prepared and then we obtained lymphatic fistulae by cutting the hypertrophic lymph-nodes situated around the external iliac vessels. These lymphatic masses did not constitute an obstructive dam which could have impaired the lymphatic drainage, but, contrarily, they were under a lymphatic stasis due to a more proximal dam: after cutting or crushing these hypertrophic lymph nodes we obtained a significant lymphoragia which proved that the lymphatic obstruction is located in a more proximal area; we did not identify any visible lymphatic structures proximally, around the common iliac vessels or adjacent to the terminal abdominal aorta. Finally, there was obtained a pelvic lymphoragic basin which was sustained by the surgically injured distal-iliac lymph-nodes.

- Connection of the "absorptive" intestinal rectangle to the rectangular pelvic lymphoragic area by continuous Polypropylene suture. Between the 2 areas (lymphoreic and absorptive) we also introduced an omental flap for its weak and short-term absorptive property and especially for its anti-infection properties. We didn't perform any microsurgical connections between this omental flap with any iliac lymphatic structure. We inserted external drain tubes inside both Scarpa triangles and in the pelvis (both inside the extra-peritoneal lymphoreic area and in the inferior intra-peritoneal cavity).

Results

After the operation the external drainage progressively and significantly diminished and we could remove the drain tubes after 12 days. The patient did not present any sign of infection. The limbs swelling disappeared and the genitalia and limb inflammatory lesions healed in

less than 20 days. 3 months after the enteromesenteric bridging operation there was resected the remnant scrotal sclero-lymphatic tissue. We didn't notice any early or late post-surgery complication. 3 years after surgery the clinical control showed no lymphedema recurrence; the perimeter measurement of the limb performed in order to evaluate clinically the loss of volume after the surgery showed a decrease of the mid-calf diameters with 5.2 cm on the right and 4.8 cm on the left (comparing with the preoperative measurements). Post-surgery the patient didn't present any recurrent limb or perineal infection and the limbs didn't present any trophic lesions. (Fig. 3). There were done no late postoperative imaging explorations because the patient refused other medical interventions.

Discussion

Primary lymphedema represents a condition which affects the lymph vessels and their cells without any pathologic patient history leading to soft tissue swelling. Usually they presents a family history with autosomal or recessive gene transmission⁶. The anatomical segments involved are usually the limbs, on one or both sides, more often the inferior ones. Congenital primary lymphedema, or Milroy disease, represents 20% of primary lymphedemas. In disease pathogenesis, we need to recall the involvement of the gene responsible for vascular endothelial growth factor receptor-3 (VEGFR-3) tyrosine kinase signaling mechanism, which is thought to be specific to lymphatic vessels and plays an important role in lymphatic development⁷. In Milroy disease, the symptoms are obvious starting from birth^{6,7}. When lymphedema occurs in puberty and develops until 35 years of age, it belongs to another type of primary lymphedema – lymphedema praecox. The symptoms involve the anatomical area situated below the waist. females are about four times more affected than men by? the most frequent type of primary lymphedema^{6,8}. In the literature, it's used the Meige disease or Meige syndrome term for both conditions, but more often it refers only to lymphedema praecox. The third type of primary lymphedema is lymphedema tarda, when, the same symptoms develop in over 35 yearold patients. It's the rarest type of primary lymphedema⁶. Despite the obvious clinical features, the positive diagnosis is sustained by lymphoscintigraphy and CT scan or MRI⁹.

The case presented belongs to the second type – lymphedema praecox (18 years old). The clinical diagnosis was sustained by simple imaging techniques (ultrasound and computed-tomography scans). Neither the imaging diagnostic techniques, nor the intraoperative features revealed anysign of mesenteric lymphatic stasis or occlusion. Having these findings, we decided to use the principle of Kinmoth's operation in which the lymphatic stasis is drained from the lower limbs and genitalia toward the mesenteric lymphatic circulation: thus, we bypassed



Fig. 3

the iliac and retroperitoneal occluded lymphatic circulation. We applied the intestinal absorptive patch on a wider lymphoragic area, which is sustained by a greater number of injured lymph-nodes (both intra-abdominal and inguinal). Therefore, a lymph drainage from a wider territory was obtained, from the lower limbs and from the perineal territory as well. As a redundant measure, we associated the omental flap transfer into the lymphoragic area in order to increase the lymphatic bypass even for a short-term after surgery⁸ and especially for its anti-infectious properties. We prevented postsurgery complications (lymphocele, a potentially huge lymph pool or a pelvic abscess) by associating some prophylactic measures: insertion of the omental flap inside the lymphoreic area, antibiotics coverage for minimum 7 days post-surgery, drain tubes both in the inguinal area until getting a significant decrease of the external drainage content. Having no experience with microsurgical techniques which demonstrated excellent long-term results⁹⁻¹¹, we choosed this lymphatic enteromesenteric bypass which was easily performed and which finalized with good long-term results and without any postoperative complications. In order to remove the excess volume of the perineal organs due to the excessive sclero-lympatic tissue we resected the fibro-lymphatic subcutaneous scrotal excessive tissues¹². This operation also finalized with good late results, without any postoperative complications.

Conclusion

The enteromesenteric bridge combined with the omental flap proved to be efficient for ensuring the lymphatic drainage in a case with proximal lymphatic occlusion. Our modified technique consists in creating a pelvic lymphoragic basin by injuring bilaterally the external iliac lymph-nodes; this measure assured the release of the lymphatic stasis both from the lower limbs and from the perineal region. The lymph basin was then efficiently drained both by the enteromesenteric bridge and by the transferred omental flap. Using this modified technique we didn't have any early or late postoperative complication and we obtained good late results, with the permanent relief of the lymphedema.

Riassunto

L'edema linfatico primario è una condizione che colpisce i vasi linfatici e le loro cellule senza alcuna storia patologica del paziente che porti a rigonfiamento dei tessuti molli. Di solito presentano una storia familiare di trasmissione di geni autosomici o recessivi. L'edema linfatico congenito, o malattia di Milroy, rappresenta il 20% degli edemi linfatici primari. Quando un edema linfatico si presenta durante la pubertà e continua a svilup-

parsi fino ai 35 anni, appartiene a una diversa categoria di edemi linfatici primari: edema linfatico precoce. I sintomi riguardano l'area anatomica al di sotto della circonferenza addominale. Nella letteratura, il termine malattia di Meige, o sindrome di Meige, si riferisce ad entrambe le condizioni, ma di solito principalmente all'edema linfatico primario precoce. Il terzo tipo di edema linfatico primario è l'edema linfatico tardivo, in cui i sintomi appaiono in pazienti al di sopra dei 35 anni di età. Quest'ultimo è la tipologia più rara di edema linfatico primario. L'operazione del ponte entero-mesenterico è considerata essere una tecnica chirurgica adatta solo per il trattamento dell'edema linfatico primario, ma nella letteratura medica i riferimenti riguardanti tale procedura sono molto rari.

Lo scopo di questo articolo è di presentare una variante della procedura di Kimmoth per il trattamento dell'edema linfatico primario precoce attraverso l'inserimento di un flap omentale all'interno delle aree di riassorbimento.

Presentiamo il caso di un paziente di 18 anni con un edema linfatico primario considerevole agli arti inferiori ed ai genitali associato con lesioni ulcerative, necrotiche ed infiammatorie nelle zone dello scroto ed al di sotto delle ginocchia. La tomografia computerizzata e la scansione ad ultrasuoni hanno mostrato linfonodi ipertrofici in entrambi i lati, sia a livello dell'area iliaca esterna che a livello dell'area inguinale, senza alcuna massa linfatica prossimale, in sede periaortica o intorno ai vasi iliaci. La linfo-scintigrafia con radionuclide non era disponibile, ma si è arrivati alla conclusione che la tomografia computerizzata associata ai sintomi clinici sia indicativa della diagnosi dell'occlusione linfatica prossimale. Alla luce della diagnosi si è deciso di intervenire con un drenaggio linfatico fisiologico, combinando il ponte entero-mesenterico con la trasposizione omentale. Abbiamo modificato tale tecnica applicandola ad un paziente con edema linfatico primario precoce dovuto ad occlusione linfatica prossimale. Al fine di bypassare l'occlusione linfatica retroperitoneale abbiamo creato un'area linforagica pelvica alla quale sono stati applicati il ponte entero-mesenterico ed un flap omentale per permettere il drenaggio della linfa. Il decorso post operatorio è stato positivo e privo di complicazioni riguardanti le ferite. Il gonfiore agli arti è scomparso e le lesioni infiammatorie ai genitali ed agli arti sono guarite in meno di 20 giorni. A distanza di tre mesi il rimanente tessuto sclero linfatico scrotale è stato rimosso. A tre anni dall'operazione il follow-up clinico non ha mostrato alcun ripresentarsi del linfedema.

CONCLUSIONE: La nostra nuova tecnica consiste nel creare un'area linforagica pelvica lesionando bilateralmente sia i linfonodi iliaci che inguinali. Tale procedura assicura lo sbloccarsi della stasi linfatica sia negli arti inferiori che nella zona perineale. Inoltre, abbiamo aggiunto un flap omentale al fine di aumentare il flusso di assorbimento della linfa. Usando questa nuova tecnica

non abbiamo riscontrato alcuna complicazione postoperatoria, né nell'immediato né a lungo termine, ed abbiamo ottenuto risultati duraturi, con la completa scomparsa dell'edema linfatico.

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Commento e Commentary

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The authors describe a combined enteromesenteric bridging and omental flap procedure applied to a case of primary lymphedema with bilateral lower limb lymphedema with genital involvement. After the creation of an 'absorptive' rectangle of small bowel, an omentum flap was placed between the rectangle of small bowel and the area of lymphorrhea in the pelvic-subperitoneal region, formed by systematic 'crushing' of the pelvic and inguinal lymph nodes, in order to improve the lymphatic drainage of the lower district. The authors report no post-operative complications and that there was a volume reduction of 10cm and 15cm for the calf and thigh respectively. The combined concept itself is interesting and improvements in the efficacy and safety of lymph node transfers is of prime importance, given the recent reports of donor lymphedema with vascularized lymph node transfers ¹.

No pre-operative imaging was performed of the lymphatic vessels and the flow, as a lymphoscintigraphy was not available. Diagnosis of primary lymphedema praecox was instead based on US and CT imaging where the inguinal and external iliac nodes were found to be hypertrophic. No post-operative imaging was completed due to loss of the patient to follow-up. Without the use of imaging, it is difficult to say whether the improved lymphatic drainage demonstrated was due to the enteromesenteric bridge, the omentum flap, or even simply to the destruction of the lymph nodes, which may have been analogous to removing the obstruction to lymphatic flow (the hypertrophic nodes). The crushing of the lymph nodes is an unusual technique, as usually the Kinmonth procedure anastomoses the enteromesenteric bridge to transected lymph nodes ^{2,3}. The current paper instead refers to simply removing a piece of the ileum and suturing it in the area of the pelvic lymphorrhea. An omental flap was also introduced in this site, presumably connected microsurgically to nearby vessels, although this is not specified by the authors. As such, the success of the surgery is difficult to evaluate as it is unclear through which means it has been achieved.

The capacity to evaluate the clinical outcome is essential to any surgery. Although the enteromesenteric bridge procedure demonstrated some initial success in a case series of patients with primary lymphedema, it has not been widely adopted, moreover due to the development of lymphatic-venous anastomosis procedures ¹. Omentum flaps were also susceptible to early fibrosis and, as such, have been modified with minimally invasive approach to overcome these limitations⁴. Furthermore, it is possible today to adequately treat peripheral lymphedema (both primary and secondary) with microsurgical techniques obtaining excellent long-term results up to 20 years after the surgery ⁵⁻⁷. For cases with advanced lymphedema and a significant fibrotic-adipose tissue component, a second properly adapted liposuction procedure with lymph vessel sparing technique can be performed to safely reduce the excess volume ⁸⁻⁹.

* * *

Gli Autori descrivono una procedura combinata di ponteggio enteromesenterico e flap omentale applicata a un caso di linfedema primario con linfedema bilaterale dell'arto inferiore e con coinvolgimento genitale. Dopo la creazione di un rettangolo "assorbente" del piccolo intestino, un lembo di omento è stato collocato tra il retto dell'intestino tenue e l'area della linforrea nella regione pelvico-subperitoneale, provocata da "schiacciamento" sistematico dei linfonodi pelvici e inguinali, al fine di migliorare il drenaggio linfatico del distretto inferiore. Gli autori non riportano complicazioni post-operatorie e una riduzione di 10 cm e 15 cm del volume rispettivamente del polpaccio e della coscia. Il concetto combinato in sé è interessante e il miglioramento dell'efficacia e della sicurezza dei trasferimenti linfonodali è di primaria importanza, date le recenti segnalazioni di linfedema del donatore con trasferimenti di linfonodi vascularizzati¹.

Non è stata eseguita alcuna imaging preoperatoria dei vasi linfatici e del flusso, in quanto non era disponibile una linfoscintigrafia. La diagnosi di linfedema primitivo praecox era invece basata su imaging US e TC in cui i linfonodi iliaci inguinali ed esterni erano risultati ipertrofici. Nessuna immagine post-operatoria è stata completata a causa della perdita del paziente al follow-up. Senza l'uso dell'imaging, è difficile dire se il miglioramento del drenaggio linfatico dimostrato fosse dovuto al ponte enteromesenterico, al lembo omentale, o anche semplicemente alla distruzione dei linfonodi, che potrebbe essere stato analogo all'eliminazione dell'ostruzione del flusso linfatico (indice i nodi ipertrofici). Lo schiacciamento dei linfonodi è una tecnica insolita, poiché di solito la procedura di Kinmonth anastomizza il ponte enteromesenterico ai linfonodi sezionati ^{2,3}. Questo studio invece riferisce semplicemente la rimozione di un frammento dell'ileo e la sua sutura all'area della linforrea pelvica. In questa sede è stato inserito anche un lembo omentale, presumibilmente collegato in modo microscopico a vasi vicini, sebbene questo non sia specificato dagli autori. In tal senso il risultato positivo dell'intervento è di difficile valutazione in quanto non è chiaro in che modo è stato raggiunto.

La capacità di valutare l'esito clinico è essenziale per qualsiasi intervento chirurgico. Sebbene la procedura del ponte enteromesenterico abbia dimostrato un successo iniziale in una serie di casi di pazienti con linfedema primario, non è stata ampiamente adottata, per lo più a causa dello sviluppo di procedure di anastomosi venosa-linfatica ¹. I

lembi omentali erano anche soggetti alla fibrosi precoce e, come tali, sono stati modificati con un approccio minimamente invasivo per superare queste limitazioni⁴. Inoltre, oggi è possibile trattare adeguatamente il linfedema periferico (sia primario che secondario) con tecniche microchirurgiche ottenendo ottimi risultati a lungo termine fino a 20 anni dopo l'intervento chirurgico⁵⁻⁷. Per i casi con linfedema avanzato e componente significativa del tessuto fibrotico-adiposo, è possibile eseguire una seconda procedura di liposuzione adeguatamente adattata con tecnica di risparmio dei vasi linfatici per ridurre in modo sicuro il volume in eccesso⁸.

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