# Laparoscopic treatment of splenic lymphangiomas: report of three cases

U. BARBAROS, U. DEVECI, Y. ÖZLÜK<sup>1</sup>, B. TIRYAKI<sup>2</sup>, Y. ERBIL, A. DINÇÇAĞ, S. MERCAN, R. VECCHIO<sup>3</sup>

SUMMARY: Laparoscopic treatment of splenic lymphangiomas: report of three cases.

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Background and aim. Since 1991, laparoscopic splenectomy has been performed in many different pathologies of the spleen. Although it is a rare lesion, splenic lymphagiomas are cystic lesions of the spleen requiring splenectomy. Herein, we present three females who have undergone laparoscopic splenectomy with the diagnosis of cystic splenic lymphangioma.

Patients and methods. In the last four years, in Istanbul Medical School, Department of General Surgery (Turkey) and in University of Catania Medical School, Department of Surgery (Italy), we performed laparoscopic splenectomy in three cases of splenic lymphangioma.

Results. These three female patients, with the age of 26, 30 and 40, had nonspecific abdominal pain requiring abdominal CT scan and magnetic resonance imaging, which showed incidental cystic lesions in the spleen, associated with cholelithiasis in one case. Preoperative laboratory tests and physical examinations were normal. Laparoscopic splenectomy was performed successfully with three 10 mm trocars in two patients in less than 1 hour, and with an Hasson trocar, two 5 mm trocars and one 10-12 mm trocar in the last case, who required simultaneous cholecystectomy. No peroperative and postoperative complications has occurred. Histopathological examinations confirmed the preoperative diagnosis.

Conclusion. Laparoscopic splenectomy is the best treatment for patients with suspected cystic lymphangioma. It permits a total pathological examination of the spleen, and it should be preferred to partial splenectomy because of possible multiple lesions. In conclusion, minimal invasive treatment of this rare pathology is an effective and safe procedure.

RIASSUNTO: Trattamento chirurgico laparoscopico di linfangiomi splenici: presentazione di tre casi clinici.

U. BARBAROS, U. DEVECI, Y. ÖZLÜK, B. TIRYAKI, Y. ERBIL, A. DINÇÇAĞ, S. MERCAN, R. VECCHI

Introduzione. Dopo la prima realizzazione nel 1991, la splenectomia laparoscopica è stata proposta ed eseguita in diverse patologie spleniche. I linfangiomi splenici rappresentano rare lesioni cistiche della milza che richiedono la splenectomia. Vengono presentati tre casi clinici di linfangiomi cistici trattati con splenectomia laparoscopica.

Pazienti e metodi. I pazienti sono stati osservati e trattati mediante splenectomia laparoscopica presso il Dipartimento di Chirurgia della Università di Instanbul e presso il Dipartimento di Chirurgia dell'Università di Catania.

Risultati. I tre pazienti, di sesso femminile e rispettivamente di 26, 30 e 40 anni, presentavano dolore addominale aspecifico. Una indagine TC ed una RMN avevano evidenziato lesioni cistiche spleniche, associate in un caso a colelitiasi. L'esame obiettivo e le indagini bioumorali eseguite pre-operatoriamente erano normali. La splenectomia laparoscopica è stata eseguita con successo con l'utilizzo di tre trocars da 10 mm in due pazienti (durata intervento 1 ora) e con un trocar di Hasson, 2 trocars da 5 mm ed 1 trocar da 10-12 mm nel terzo caso, dove è stata associata una colecistectomia. Non sono state segnalate complicanze e mortalità. L'esame istologico ha confermato la diagnosi di linfangiomi.

Conclusioni. La splenectomia laparoscopica rappresenta la modalità migliore di trattamento per i pazienti con sospetto linfangioma cistico della milza. Consente una rimozione completa della milza, da preferire alla splenectomia parziale, dal momento che tale patologia splenica può essere multicentrica. In conclusione, il trattamento miniinvasivo laparoscopico dei linfangiomi splenici è efficace e rappresenta una procedura sicura.

KEY WORDS: Spleen - Laparoscopy - Cystic lymphangioma. Milza - Laparoscopia - Linfangioma cistico.

Istanbul University, Istanbul, Turkey Medical School, Department of General Surgery Department of Pathology Department of Radiology

Medical School, Department of Surgery

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## Introduction

Cystic lymphangiomas are rare, congenital malformations of the lymphatics which are found predominantly in children (1). Most lymphangiomas locate in

<sup>&</sup>lt;sup>3</sup> University of Catania, Italy

the head and neck region; intra-abdominal locations, such as in the omentum, mesentery and retroperitoneum, occurring less commonly. They have a wide spectrum of clinical and laboratory findings, from an incidental finding to a large symptomatic mass (1). Their prognosis is good but there is a remarkable high risk of splenic rupture. Surgery is always the preferable definitive treatment (1).

We report three cases of splenic lymphangioma, and discuss both diagnostic and therapeutic aspects of laparoscopic splenectomy which is the more effective procedure in these cases.

### **Clinical series**

### Case n. 1

A 26 year old female was found to have a splenic cystic tumor during the follow-up for previous treated pulmonary tuberculosis. The physical examination was unremarkable and the laboratory findings were all within the normal limits.

Ultrasonography (US) showed multiple cystic lesions with the largest up to 3 cm in diameter. CT scan showed an image similar to that on US. Magnetic resonance imaging (MRI) revealed a low-intensity mass on T1-weighted images and a high-intensity mass on T2-weighted images (Fig. 1). Laparoscopic splenectomy was planned for diagnosis and treatment of the lesion.

Patient was placed in right semilateral position (45°) with the left side up and a flank cushion placed under the right side. Three 10 mm trocars were used. Laparoscopic exploration demonstrated splenic cysts in normal splenic parenchyma (Fig. 2). The first step was the dissection with the use of Ligasure<sup>®</sup> (Valleylab, Boulder, CO, USA) of the lower pole of the spleen and of the spleno-renal ligament. Short gastric vessels were then ligated and divided. The hilar splenic vessels also were ligated and divided with Ligasure<sup>®</sup> just before the dissection of the diaphragmatic attachment of the spleen. During the division of the splenic hilar vessels, Ligasure<sup>®</sup> probe was used as close as possible to the splenic parenchyma, far away from pancreatic tail, in order to avoid injuries. At the end the spleen was removed after its fragmentation within a special retrieval bag. The operative time and the estimated blood loss were 60 minutes and 90 ml, respectively.

The patient was discharged on the second postoperative day and followed-up in a six month period. No complications were observed. Histopathologic examination revealed splenic lymphangioma.

#### Case n. 2

A 30 year old female was found to have a splenic cystic tumor discovered during a diagnostic work-up performed for the complaint of abdominal pain. The physical examination was unremarkable and the laboratory findings were all within the normal range.

On CT scan, a 4.5 cm splenic cyst located on the lower splenic pole was defined. CT images of the lesion were consistent with the diagnosis of lymphangioma.

Laparoscopic splenectomy was planned and was performed following the same technique used in the first case described above. Laparoscopic exploration demonstrated a splenic cyst located at the lower pole of the spleen (Fig. 3). The operative time and the estimated blood loss were 55 minutes and 100 ml, respectively.



Fig. 1 - MRI view of the splenic lymphangioma.



Fig. 2 - Peroperative view of the hilar cyst covered with normal splenic parenchyma when the spleen was turned up-side down after splenectomy ended.



Fig. 3 - Peroperative view of the splenic cyst located at the lower pole.

The patients was discharged on the second postoperative day and followed-up for 5 months without the occurrence of any complication. Histopathologic examination revealed splenic cystic lymphangioma, according to the preoperative suspicion.

#### Case n. 3

A 40 year old female who was admitted to the hospital for symptomatic cholelithiasis, showed at ultrasonography a cystic lesion in the spleen in addition to the biliary disease. CT scan confirmed the single cyst of the spleen which was 6 cm in diameter in the lower pole. Laparoscopic surgery was scheduled for both gallbladder and splenic diseases.

After the insertion of an Hasson trocar in the peri-umbilical area, additional 5 mm trocar was placed in the subxiphoid region. A 10-12 mm trocar was then placed in the left flank and another 5 mm trocar was positioned in the subcostal area in the anterior axillary line. Cholecystectomy was first perfomed introducing the camera in the 10-12 mm trocar and using as working ports the Hasson trocar and the subxiphoid trocar. Afterwards, splenectomy was accomplished lifting up the left flank of the patient. Short gastric vessels were divided with the use of Ultracision® (Johnson & Johnson, Gateway, USA) and then spleno-colic and spleno-diaphragmatic ligaments were dissected. Splenic hilum was then divided away from the pancreatic tail using endovascular linear stapler. Spleen was removed within a large endobag after its fragmentation. The operation lasted about 90 minutes and blood loss was less than 50 ml.

Patient was discharged from the hospital in the second postoperative day. No complication were observed in one year followup. Pathological diagnosis was splenic lymphangioma and chronic cholecystitis.

### Discussion

Lymphangiomas are usually described as benign neoplasms but should probably be considered malformations made of abnormally dilated lymphatic vessels. Lymphangiomas are usually divided into three types - capillary, cavernous and cystic - depending on the size of the dilated lymphatic channels (1).

Abdominal lymphangiomas result from developmental failure of the lymphatics and have recently been differentiated from mesenteric cysts, which originate from mesothelial tissue (2). Lymphangiomas can be characterized pathologically by a flat epithelial endothelium and a wall containing alternatively lymphoid tissue, small lymphatic spaces, smooth muscle, and foam cells (2). Brian at al. hypothesize that endogenous estrogens may also play a role in the enlargement or growth of lymphangiomas thus explaining the female preponderance of intra-abdominal lymphangiomas in adults as compared to children.

Splenic lymphangiomas are rare and only occasional reports have been published in the literature (1, 3, 4). The most common type of presentation of splenic lymphangiomas is pain caused by pressure of the cyst on adjacent viscera. Other symptoms, such as anorexia, nausea, vomiting, or symptoms related to displacement and pressure on the stomach or splenic flexure of the colon, are not common. Physical examination may be normal or may reveal a left upper quadrant mass.

In some cases, like our cases, cystic lymphangiomas are asymptomatic and are discovered incidentally. At US the cyst are anechoic or hypoechoic with internal debris. Calcifications may be detected if they cause acoustic shadowing. On CT scan the appearance is that of a mass made up of multiple low density thin walled sharply marginated cysts which may contain mural calcifications. After intravenous injection of iodinated contrast the cystic fluid does not enhance, while the septations may present moderate enhancement (3). On MRI the mass is shown as multiloculated hyperintensity areas on the T2-weighted images, thus corresponding to the dilated lymphatic spaces. The advantage of MRI is that it is multiplanar and multiparametric; this is important for the identification of possible areas of malignant degeneration (3).

The treatment consists of surgical removal of the spleen. In 2001, Kwon et al. presented the first case report of a successful laparoscopic complete excision of a splenic lymphangioma (4). Several reports have confirmed the advantages of minimal invasive splenectomy, and these include decreased pain, lower morbidity and transfusion rate, less postoperative ileus, shorter hospital stay, earlier return to full activity, and improved cosmesis when compared with conventional splenectomy. Also our patients discharged on the second postoperative day without any complication.

Although total splenectomy is the most common treatment modality for lymphangiomatosis, other treatment options have been described. Percutaneous aspiration under ultrasound or CT scan guidance has been used but it seems of little value (5). Lymphangiomas are sometimes multifocal and leaving the diffusely involved spleen may result in recurrence of the lesions with the need of further treatments. For the same reasons we thought that partial splenectomy is not appropriate for this disease of the spleen. Although the majority of splenic lymphangiomas are benign, total splenectomy seems, therefore, to be the preferred treatment choice in order to avoid recurrence and to remove lesions whose malignancy cannot be ruled out preoperatively.

Splenectomy performed by laparoscopy is currently the preferred surgical approach in referred centers. In conclusion, it can be declared that minimal invasive treatment of this rare pathology is effective and safe procedure. U. Barbaros et al.

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