Endoscopy International Open

EUS guided Endoscopic Internal Drainage with Lumen Apposed Metal Stent of symptomatic hepatic cysts. A case series (with video)

Francesca D’Errico, Serge Derhy, Maurizio Fazi, Riccardo Memeo, Francesco Decembrino, Giovanni D. De Palma, Gianfranco Donatelli.

Affiliations below.

DOI: 10.1055/a-1968-7596

Please cite this article as: D’Errico F, Derhy S, Fazi M et al. EUS guided Endoscopic Internal Drainage with Lumen Apposed Metal Stent of symptomatic hepatic cysts. A case series (with video). Endoscopy International Open 2022. doi: 10.1055/a-1968-7596

Conflict of Interest: The authors declare that they have no conflict of interest.

Abstract:
INTRODUCTION/AIM: Simple hepatic cysts (SHC) are usually asymptomatic and detected incidentally. However, larger cysts may present with clinical signs and require treatment such as percutaneous aspiration or surgery with non negligible rate of recurrence. We report a series of 13 consecutive patients who underwent EUS guided Lumen Apposed Metal Stent (LAMS) drainage of symptomatic hepatic cysts of the right and left liver.
METHODS: 9 Males, 4 Female, average age of 71.9 years underwent EUS guided LAMS cyst drainage because of significant symptoms. At one month, LAMS was changed for double pigtail stent (DPS) for three months. 9 were located in the right liver and four in the left. Diameter was on average 22.2 cm.
RESULTS: 13 LAMS were successful delivered in all patients. However only in 12 out of 13 (92.3%) remained in place. In one case it slipped out immediately, was promptly removed and the cyst treated percutaneously. 1 out 12 presented bleeding and were treated conservatively. Only in 7 patients LAMS was removed for DPS, in the other 5 were successfully left in place until death given their comorbidities. At 10.5 months of follow up none recurred.
CONCLUSION: EUS guided LAMS drainage permits treatment of symptomatic hepatic cysts without recurrence and with few adverse events. Studies of comparison are needed to consider this approach as first intention.

Corresponding Author:
MD Gianfranco Donatelli, Hôpital Privé des Peupliers, Générale de Santé, Unité d’Endoscopie Thérapeutique, Paris, France, donatelli@unina.it

Affiliations:
Francesca D’Errico, “F. Miulli” General Regional Hospital, Acquaviva delle Fonti, Unit of Gastroenterology and Digestive Endoscopy, Acquaviva delle Fonti, Italy
Serge Derhy, Hôpital Privé des Peupliers, Unité de Radiologie Interventionnelle, Paris, France
Maurizio Fazi, Hôpital Privé des Peupliers, Générale de Santé, Unité d’Endoscopie Thérapeutique, Paris, France
Gianfranco Donatelli, University of Naples Federico II, Department of Clinical Medicine and Surgery, Napoli, Italy

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.
EUS guided Endoscopic Internal Drainage with Lumen Apposed Metal Stent of symptomatic hepatic cysts. A case series (with video)

Francesca D’Errico 1,2 MD, Serge Derhy 1 MD, Maurizio Fazi 1 MD, Riccardo Memeo 3 MD, Francesco Decembrino 2 MD, Giovanni D. De Palma4 MD, Gianfranco DONATELLI 1,4 MD.

1 Unité d’Endoscopie Interventionnelle, Hôpital Privé des Peupliers, Ramsay Santé, Paris, France
2 Unit of Gastroenterology and Digestive Endoscopy,"F. Miulli" General Regional Hospital, Acquaviva delle Fonti
Corresponding author:

Prof. Gianfranco DONATELLI,

Unité d’Endoscopie Interventionnelle,

Ramsay Santé, Hôpital Privé des Peupliers

8 Place de l’Abbé G. Hénocque

75013, Paris, France

Phone +33 6 64 95 00 70

Fax + 33 01 44 16 56 15

Email: donatelligianfranco@gmail.com
gianfranco.donatelli@unina.it

ABSTRACT
INTRODUCTION/AIM: Simple hepatic cysts (SHC) are usually asymptomatic and detected incidentally. However, larger cysts may present with clinical signs and require treatment such as percutaneous aspiration or surgery with non negligible rate of recurrence. We report a series of 13 consecutive patients who underwent EUS guided Lumen Apposed Metal Stent (LAMS) drainage of symptomatic hepatic cysts of the right and left liver.

METHODS: 9 Males, 4 Female, average age of 71.9 years underwent EUS guided LAMS cyst drainage because of significant symptoms. At one month, LAMS was changed for double pigtails stent (DPS) for three months. 9 were located in the right liver and four in the left. Diameter was on average 22.2 cm.

RESULTS: 13 LAMS were successful delivered in all patients. However only in 12 out of 13 (92.3%) remained in place. In one case it slipped out immediately, was promptly removed and the cyst treated percutaneously. 1 out 12 presented bleeding and were treated conservatively. Only in 7 patients LAMS was removed for DPS, in the other 5 were successfully left in place until death given their comorbidities. At 10.5 months of follow up none recurred.

CONCLUSION: EUS guided LAMS drainage permits treatment of symptomatic hepatic cysts without recurrence and with few adverse events. Studies of comparison are needed to consider this approach as first intention.
INTRODUCTION

Simple hepatic cysts (SHC) are the most commonly diagnosed benign liver lesions, with a prevalence of 18% among the general population undergoing abdominal CT imaging for unrelated pathologies [1]. They are usually asymptomatic and detected incidentally given the development and wide spread use of diagnostic modalities [2]. However, larger lesions may present with clinical signs such as abdominal pain, epigastric fullness, early satiety or even jaundice. Infrequently, internal hemorrhage, infection, or rapid enlargement can lead to symptoms and presentation for clinical evaluation [2]. Asymptomatic simple hepatic cysts do not require treatment. In contrast, symptomatic simple hepatic cysts might be considered for percutaneous aspiration, aspiration followed by sclerotherapy or surgery [3].

EUS guided Endoscopic Internal Drainage (EID) could be an attractive minimally invasive alternative to the percutaneous one [4]. In addition, recent development of dedicated lumen apposing metal stents (LAMS) mounted on an electrocautery enhanced delivery system, made possible a one-step procedure with formation of a stable communication between the gastrointestinal (GI) lumen and the cystic abdominal cavity [5].

Here we report the first experience, to our knowledge, of 13 consecutive patients who underwent EUS guided LAMS drainage of symptomatic hepatic cysts of the right and left liver.
MATERIALS AND METHODS

Since January 2019 all patients with symptomatic hepatic cysts of the right or left liver lobe, never treated or recurred after surgical or percutaneous drainage, were treated with EUS guided LAMS drainage (video 1) and included in a database. Data were retrospectively analyzed. Protocol was approved by ethic’s local hospital medical commission. As per protocol, LAMS was left in place for 4 weeks then changed with double pigtail stent (DPS) (video 2) and the latter were withdrawn three months later. Thirteen patients (9 M) with an average age of 71.9 years (45-98) were included (table 1).

All patients presented with pain and vomiting; in addition, six of them presented with several sepsis episodes, two had jaundice due to the compression of the cyst on the biliary tree and one developed pancreatitis. Three patients presented with ascites. Moreover, three patients presented with compression on the portal vein with thrombosis and they were treated with anticoagulant therapy. All of them were critically ill, presenting with severe malnutrition and sepsis at the time of endoscopic intervention. Cardiac failure with significant reduction of the ejection fraction was present in 3 of them. In all patients EUS guided EID LAMS drainage was performed.
None of the patients enrolled in the protocol had a history of hepato-biliary endoscopy, surgery or trauma. Five patients presented with cyst recurrence, three after percutaneous drainage at a time span of 90 days (15–120 days) and two after surgical fenestration at 8 months and 1 year respectively. Nine cysts developed from the right liver lobe (Fig.1a,b), four of them from the left one. On average, the size of the cysts treated was 22.2 cm and the biggest was 40 cm (11–40 cm). They were unilocular with mostly liquid content with about 30% of the total volume corpuscular and thick. A preoperative CT scan was performed in all patients as well as a post-operative one at 1 week and 3 weeks after LAMS placement. No signs of malignancies neither of a hydatid origin were identified. LAMS used was Hot Axios (Boston Scientific, Massachusetts, USA), two 20x10 mm, nine 15x10 mm and two 10x10 mm. The technique used was the free hand one. Once the cyst was identified, the best position was found to have the less tissue interposition, avoiding vessels and punching the cyst in its lowest part, trying to maximize, in this way, its emptying. A trans-esophageal approach was used in 5 cases, trans-gastric in 5 and trans duodenal in 3 of them. Fluoroscopy was not used in all cases. In all patients, aspiration of liquid was performed for cultural and biochemical examinations.

RESULTS:
Technical success was defined as EUS identification through the upper GI tract (oesophagus, stomach, duodenum) and puncture of the cyst with deployment of LAMS and release of liquid in the GI tract allowing to perform bacteriology examinations. This was achieved in 92% of cases (12 out 13). In one patient (number 2 of the series), who underwent a trans-oesophageal drainage for a huge left liver cyst, after the deployment of LAMS and aspiration of the liquid for bacteriology, the proximal oesophageal flange slipped out into the peritoneal cavity between the cardias and the left liver cyst. Promptly, the access cautery defect was catheterised (Natural Orifice Transluminal Endoscopic Surgery) which would allow for the LAMS anchored in the hepatic parenchyma (cyst) to be found, grasped and pulled with the aim of repositioning it. However, this was technically difficult, so the decision was made to remove it and to close the oesophageal defect with an Ovesco clip. Immediate radiology drainage was performed with recurrence seven months later and then another EUS guided LAMS drainage was performed, successfully. Of note, this second drainage was not included in the series.
The drainage was successful in all other 12 patients (100%) with discharge from the hospital within the next few days with normal diet. All patients underwent a CT scan 7 to 21 days after the drainage and they were scheduled for LAMS removal at four weeks. In all of the twelve patients symptoms improved as well as sepsis resolved (video 2), however only seven patients (58.3%) underwent an endoscopic re-look in order to remove LAMS and replace it with double pigtail stents (Fig 2a,b) for three months (Fig3a,b). For the other five patients, considering their age, co-morbidities and successful recovery post procedure, LAMS was left in place, until death, with an average follow-up of 9 months without adverse events or inflammatory syndrome. At an average of 10,5 months (2-20) from EUS guided LAMS drainage none of the patients presented recurrence, including those with LAMS left in place (6,6; 2-13). In none of the patients a necrosectomy or debridement was needed; however it was always possible to enter in the cystic cavity through the LAMS at 1 month control (video 3). Only 1 patient, three weeks after index EUS, presented melena with anemia without active bleeding at CT scan, needing blood transfusion. LAMS was changed for pigtail stent as scheduled, at four weeks. Follow up was uneventful.

DISCUSSION
Simple hepatic cysts are typically saccular, thin-walled masses with fluid-filled epithelial lined cavities. They arise from aberrant bile duct cells that originate during embryonic development. Simple hepatic cysts are usually <1 cm and can grow up to 30 cm [7].

While they are generally incidentally diagnosed and mainly asymptomatic, symptomatic hepatic cysts need to be treated. The American College of Gastroenterology Clinical Guidelines recommend laparoscopic fenestration to deroof a cyst with or without omentoplasty as first treatment approach because of its high success rate and low invasiveness [3], however the recurrence rate is 15-20% [8]. Other techniques are described such as percutaneous drainage, puncture and sclerosis and/or injection of ethanol, with the same issue of recurrence as the surgical approach [8].

In detail, percutaneous procedures for treatment of simple hepatic cysts are particularly effective for immediate palliation of patients’ symptoms; however, they are not generally resolutive long term because of the high rate of recurrence (9) and often complicated by adverse events such as leak, skin inflammation and accidental withdrawn of the drain. Radical surgical cyst excision seems to be curative without recurrence but often characterized by a significant rate of morbidity and mortality [8].
LAMS, originally designed for drainage of transmural pancreatic fluid collections, in the last few years have been used extensively for other indications. In fact, more recently other in- and off-label indications have been proposed [6], allowing nowadays the drainage of almost all types of intra-abdominal (upper and lower GI) collections and even to perform EUS guided gastrointestinal anastomosis [10,11], with very low rate of morbidity and mortality. Despite their worldwide use there are few reports of LAMS-EUS guided drainage of symptomatic simple hepatic cysts [12], all of them performed on cysts located within the left liver lobe.

The choice of using LAMS as first intention, despite clear contents without debris, is related to avoid leak between the stomach and liver (cyst). With breathing movements, the liver and the stomach are mobile so, using pigtails as first intention which guarantee less stability than LAMS, could easily the cause a leak at the level of their insertion with a risk of peritonitis. LAMS have been developed in order to avoid this kind of adverse events as in digestive anastomosis. Once the fistula between the stomach and the liver cyst is epithelized (one month later) pigtail are useful to keep the internal drainage open, acting also as foreign bodies to promote granulation tissue formation and healing of the cyst (collapse of cavity and weld of wall).
However, abscess drainage [13] of the right liver lobe has already been attempted. Based on our experience, as long as the target is identified with EUS at a distance from the GI wall of 1cm maximum, it is always possible to proceed with a LAMS guided procedure (anastomosis or drainage). In detail, the right liver is visualized trans-gastrically in pushed position or trans-bulbarly and, despite the difficult location, generally the cysts are easily drainable due to their size which cause their walls to be attached the GI one. Infected cysts are the most common candidates for this kind of drainage given they can reach a size of more than 10 cm, increasing the likelihood of exclusion symptoms in adjacent organs [14] and making their treatment necessary. However, some concerns for EUS guided liver cysts drainage persist mainly in regard to the risk of recurrence.

In this field, antibiotic treatment only has been shown to have a treatment success rate of 20%, which, if combined with percutaneous drainage, goes up to 65%. Interestingly, antibiotics with surgery has a success rate of 100% with an overall recurrence of 20% [15]. In this consecutive series of patients, we had 100% of success coupling antibiotics with LAMS drainage followed by pigtails positioning with no clinical recurrence at an average follow up of 10.5 months, achieved with close monitoring of patients’ conditions after the procedure.
Our case series shows that persistent drainage, facilitated by the positioning of double pig-tail stents after LAMS removal, is enough to have secondary collapse of pseudo-cavity as shown in case of collection after EID of leaks and fistula after surgery, successfully treated with double pigtail stents [16]. The cause of recurrence after surgical deroofing, especially for cysts located below the diaphragm, seems to be its partial occlusion by muscles during breath movements [8]. In our technique, lowest and continuous drainage, facilitated also by pigtail positioning, leads to collapse of the pseudocavity and sticks to the pseudo-walls. Certainly, the large caliber of LAMS allows an effective drainage of cysts even if there is some necrosis such as in the walled off necrosis[17]. This is also the reason why EID by LAMS works better than percutaneous drainage for which usually the caliber of the drains used is 10-12 F. Considering also the rate of drain’s adverse events, LAMS drainage is becoming, especially in unfit patients, the preferred option for gallbladder drainage [9]. On the other hand, the massive emptying of the cyst in the GI cavity during the LAMS procedure (video) carries a potential oro-tracheal inhalation risk of the cystic content therefore it needs to be performed carefully and in experts’ hands.
Interestingly, we reported just one failure (7.7%), in a patient who needed drainage of a left liver cyst. The approach was through the intra-abdominal lower esophagus which was probably the main reason for the failure. After a hand free puncture and fully deployment of LAMS (pictures) with initial emptying of the cyst, because of breathing movements, the proximal flange slipped into the abdominal cavity causing a stent migration. In this case and previous one (the first) we use the 10x10 mm LAMS (despite the huge size of cyst, with pure liquid content) which maybe favored the migration given the cyst size. In all other patients we used at least 15mm LAMS to obtain a better anchoring, as we do routinely in the EUS guided gastro-entero anastomosis, and we did not experience any adverse events even for trans esophageal drainage.

CONCLUSION
EUS guided LAMS drainage permits to solve symptomatic hepatic cysts without recurrence and with very few adverse events. Studies of comparison with interventional radiology and surgical management are necessary to consider this approach as first intention.
Photos/video:

1a-b: CT scan showing right liver cyst, more than 20 cm of diameter
2 a-b: RX showing LAMS in place, at 1 month, changed with double pigtails
3a: Endoscopic appearance of trans gastric pigtail in place at 3 months
3b: Upper swallow study through the scope showing the absence of extravasation of medium contrast meaning that the cyst is completely resolved.

At Xray the cavity is not visible anymore at the level of the pigtails.

Videos 1: EUS guided LAMS drainage without fluoroscopy of a huge hepatic cyst with release of clear liquid.
Video 2: Abdominal CT scan at 3 months showing complete healing of the cyst
Video 3: Endoscopic exploration of the cyst at 1 month and change of LAMS with double pigtail
BIBLIOGRAPHY


9) Erdogan D, van Delden OM, Rauws EA, et al. Results of percutaneous sclerotherapy and surgical treatment in patients with symptomatic simple liver


1a-b: CT scan showing right liver cyst, more than 20 cm of diameter

2 a-b: RX showing LAMS in place, at 1 month, changed with double pigtails

3a: Endoscopic appearance of trans gastric pigtail in place at 3 months

3b: Upper swallow study through the scope showing the absence of extravasation of medium contrast meaning that the cyst is completely resolved.

At Xray the cavity is not visible anymore at the level of the pigtails.

Videos 1: EUS guided LAMS drainage without fluoroscopy of a huge hepatic cyst with release of clear liquid.

Video 2: Abdominal CT scan at 3 months showing complete healing of the cyst

Video 3: Endoscopic exploration of the cyst at 1 month and change of LAMS with double pigtail
**Table 1: Demographics data and results**

<table>
<thead>
<tr>
<th>Number</th>
<th>Sex (M/F)</th>
<th>Age (y)</th>
<th>Localization</th>
<th>Diameter (cm)</th>
<th>Previous Treatment (Yes/No - Surgical/Radiological)</th>
<th>EUS LAMS Success (Yes/No)</th>
<th>Withdrawn of LAMS (Y/N)</th>
<th>Follow-Up (Months)</th>
<th>Clinical Success (Yes/No)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>6</td>
<td>R</td>
<td>11</td>
<td>N</td>
<td>Y</td>
<td>Y</td>
<td>Y</td>
<td>Y</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>5</td>
<td>L</td>
<td>18</td>
<td>N</td>
<td>N</td>
<td>Y</td>
<td>N/A</td>
<td>N/A</td>
</tr>
<tr>
<td>3</td>
<td>M</td>
<td>8</td>
<td>L</td>
<td>13</td>
<td>Y-S</td>
<td>Y</td>
<td>N</td>
<td>13</td>
<td>Y</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>4</td>
<td>R</td>
<td>16</td>
<td>Y-R</td>
<td>Y</td>
<td>Y</td>
<td>9</td>
<td>Y</td>
</tr>
<tr>
<td>5</td>
<td>M</td>
<td>6</td>
<td>R</td>
<td>31</td>
<td>Y-R</td>
<td>Y</td>
<td>Y</td>
<td>21</td>
<td>Y</td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>6</td>
<td>R</td>
<td>19</td>
<td>N</td>
<td>Y</td>
<td>15</td>
<td>Y</td>
<td>12</td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>8</td>
<td>R</td>
<td>20</td>
<td>N</td>
<td>Y</td>
<td>15</td>
<td>N</td>
<td>8</td>
</tr>
<tr>
<td>8</td>
<td>M</td>
<td>7</td>
<td>L</td>
<td>17</td>
<td>Y-S</td>
<td>Y</td>
<td>15</td>
<td>Y</td>
<td>20</td>
</tr>
<tr>
<td>9</td>
<td>M</td>
<td>9</td>
<td>R</td>
<td>25</td>
<td>N</td>
<td>Y</td>
<td>15</td>
<td>N</td>
<td>2</td>
</tr>
<tr>
<td>10</td>
<td>M</td>
<td>9</td>
<td>R</td>
<td>40</td>
<td>Y-R</td>
<td>Y</td>
<td>20</td>
<td>N</td>
<td>7</td>
</tr>
<tr>
<td>11</td>
<td>M</td>
<td>4</td>
<td>R</td>
<td>18</td>
<td>N</td>
<td>Y</td>
<td>15</td>
<td>Y</td>
<td>5</td>
</tr>
<tr>
<td>12</td>
<td>M</td>
<td>8</td>
<td>L</td>
<td>33</td>
<td>N</td>
<td>Y</td>
<td>20</td>
<td>N</td>
<td>3</td>
</tr>
<tr>
<td>13</td>
<td>F</td>
<td>7</td>
<td>R</td>
<td>28</td>
<td>N</td>
<td>Y</td>
<td>15</td>
<td>Y</td>
<td>16</td>
</tr>
</tbody>
</table>