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Case Report

Laparoscopic Liver Resection with a SAND Balloon Catheter for a Large Mucinous Cystic Neoplasm of the Liver

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ABSTRACT: A 48-year-old woman was referred to our department for managing the regrowth of a cystic lesion of the liver. MRI revealed an 11-cm multilocular cystic lesion occupying the left lobe. Given the lack of obvious malignant findings such as intracystic nodules, we decided to perform laparoscopic liver resection (LLR). During the surgery, retracting the large hepatic cystic lesion proved difficult. We therefore aspirated the cystic contents using a SAND balloon catheter, after which the cyst wall could be managed. Ultimately, LLR could be performed fully after sequentially dissecting the Glissonean vessels toward the cystic wall. Given the presence of ovarian-like stroma below the cystic epithelium, the patient was diagnosed with mucinous cystadenoma of the liver with low-grade dysplasia. For the patient with a large mucinous cystic neoplasm of the liver, a SAND balloon catheter may prevent the leakage of cystic contents and is considered an effective device for facilitating LLR.

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KEYWORDS: mucinous cystic neoplasm of the liver, laparoscopic liver resection, SAND balloon catheter

Introduction

Mucinous cystic neoplasm of the liver (MCN-L) is a rare lesion, accounting for fewer than 5% of all cystic liver tumors.¹⁾ In recent years, laparoscopic liver resection (LLR) has been widely performed for liver tumors. Especially for low-grade malignant liver tumors, LLR is considered a beneficial surgical procedure due to its smaller incision, less pain, and superior cosmetic results. However, the use of LLR in large MCN-L may cause cystic rupture due to

the intraoperative laparoscopic procedure. Therefore, open hepatectomy may be desirable to prevent cystic rupture during surgery. If the patient strongly desired LLR, an appropriate intraoperative management is therefore required to prevent the leakage of cystic contents.

Herein, we report a case of a large MCN-L in which we were able to prevent cystic rupture and complete the LLR using a SAND balloon catheter.

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Case Report

A 48-year-old Japanese woman was diagnosed with a large symptomatic hepatic cyst 23 years earlier and underwent laparoscopic hepatic cystic fenestration. The pathological examination of the resected specimen led to the diagnosis of ectopic endometriosis. Ten years later, regrowth of the hepatic cyst was once again observed, and the patient once again underwent laparoscopic hepatic cystic fenestration. Pathological examination of the resected specimen led to the diagnosis of mucinous cystadenoma of the liver with low-grade dysplasia. This time, regrowth of the hepatic cyst was observed, and the patient was referred to our department for further examination and treatment.

Blood tests showed a slight increase in tumor markers (cancer antigen 19-9 at 94.5 U/mL). In terms of imaging, only plain CT and MRI and contrast-enhanced ultrasound were performed because of the patient's allergy to iodine contrast medium. MRI revealed an 11-cm multilocular cystic lesion occupying the left lobe of the liver (Fig. 1). The entire left lobe of the liver was occupied by cystic lesions, and the left hepatic Glissonean pedicle was not observed. The cystic lesion was in close contact with the right anterior Glissonean pedicle from the hepatic hilum. Ultrasound findings showed bleeding and necrotic material in some of the cysts, and the contrast-enhanced findings of the cyst wall led to the diagnosis of MCN-L recurrence. Given that no intracystic nodule was observed, we judged that there was no obvious malignant lesion.

Even with no obvious malignant findings, the leakage of cystic contents during surgery may lead to postoperative disseminated recurrence. From this point of view, we initially proposed extended left hepatectomy or left trisegmentectomy of the liver with caudate lobectomy by open surgery for this patient. However, MCN-L has a lower malignancy than pancreatic MCN,¹⁾ and there were no obvious malignant findings on the images. Therefore, these operative procedures were considered over-surgery. In the case of MCN-L with a low-grade dysplasia as in the previous surgery, postoperative recurrence could be prevented by removing the entire cystic lesion.²⁾ As a result, we proposed entire left hepatic cystectomy, which preserved the anterior Glissonean pedicle, hepatic hilum, and caudate lobe. The patient was given multiple explanations regarding the surgical procedure. However, because this patient had previously undergone two laparoscopic surgeries,

open surgery was refused and laparoscopic surgery was strongly desired.

Laparoscopic surgery for large cystic tumors may result in the leakage of cystic contents due to unexpected cyst rupture during surgery,³⁾ and it is difficult to sufficiently aspirate the cystic contents during the surgery. Therefore, referring to the report by Nakamura et al.,⁴⁾ using a SAND balloon catheter (Hakko Shoji, Tokyo, Japan) for the large pancreatic MCN, we aspirated the cystic contents in advance using the SAND balloon catheter in our case.

Six-port laparoscopic surgery was started under the pneumoperitoneum. Owing to the difficulty in retracting the large hepatic cystic lesion (Fig. 2A), we aspirated the cystic contents using a SAND balloon catheter (Fig. 2B). We performed cystic aspiration in four locations. The cyst puncture site was sutured laparoscopically. During these procedures, there was no leakage of cystic contents under laparoscopic view during the puncture, aspiration, and suture of the puncture site. There were also no obvious malignant findings as a result of the intraoperative cytology. After the aspiration of the cystic contents (Fig. 2C), the cyst wall could be retracted using atraumatic forceps as carefully and gently as possible. Obvious left hepatic Glissonean was not observed because of the large cystic lesion. Therefore, sequentially dissecting the Glissonean vessels toward the cyst wall was performed. The middle hepatic vein was preserved, and the left hepatic vein was divided around the hepatic parenchyma using a linear stapler. LLR could be performed completely with the entire cystic wall (Fig. 2D).

The resected specimen demonstrated a 13 × 8 × 4.5 cm multiloculated cyst (Fig. 3A, B). Histological examination revealed that the cyst wall was composed of columnar epithelium with intracellular mucus. Low papillary growth was observed in certain areas, but there were no findings suggestive of malignancy (Fig. 3C, D). Given the presence of ovarian-like stroma below the cystic epithelium, the patient was diagnosed with mucinous cystadenoma of the liver with low-grade dysplasia as before.

After surgery, a transient gastric emptying disorder was observed. However, the condition improved with conservative treatment, and the patient was discharged without any biliary complication on postoperative day 14. Eighteen months after surgery, the patient visited the outpatient clinic with no recurrence or problems.

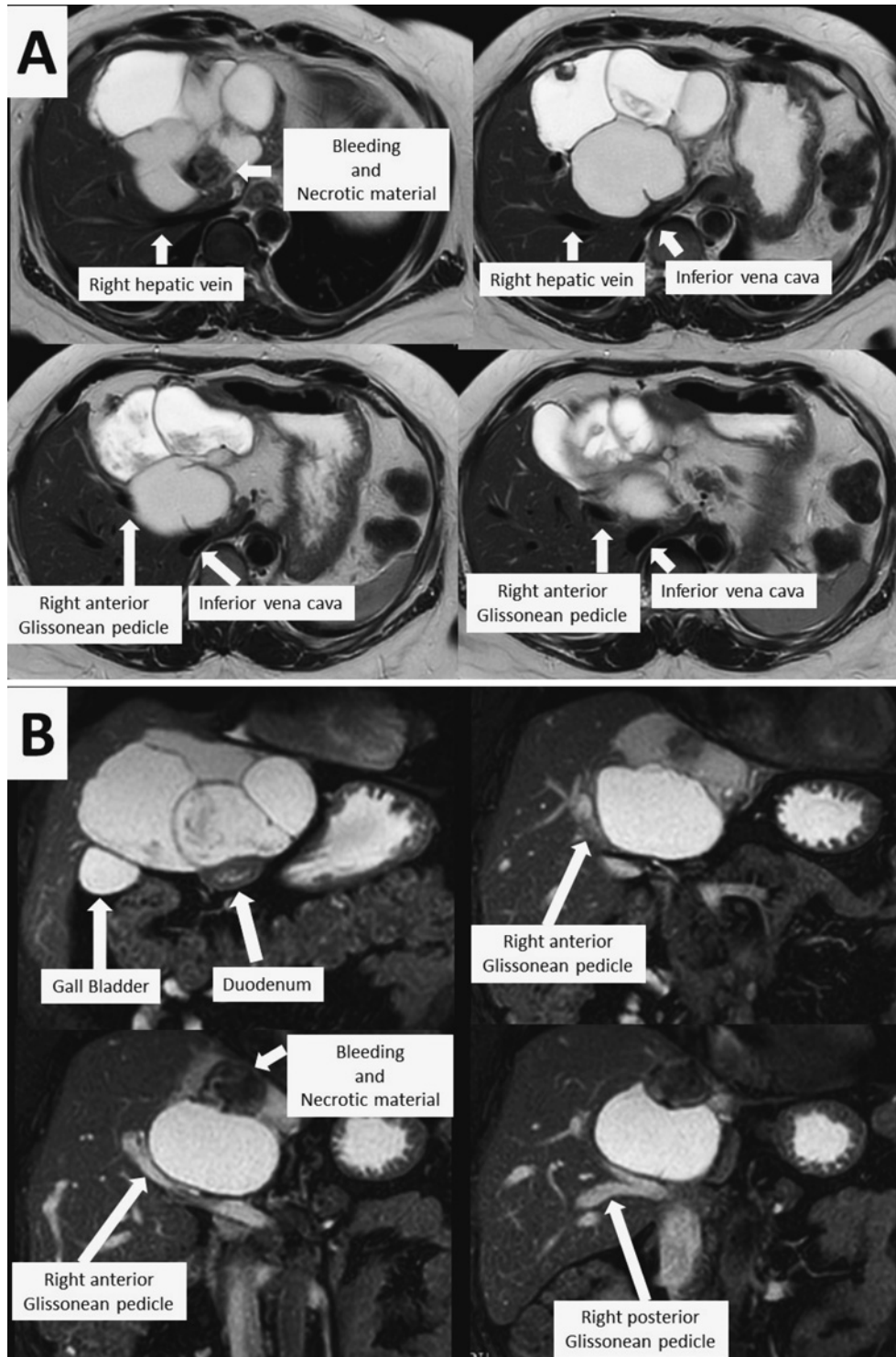


Fig. 1 MRI findings

MRI revealed an 11-cm multilocular cystic lesion occupying the left lobe of the liver. (A: axial view, B: coronal view)

Discussion

To the best of our knowledge, this is the first report in which a large MCN-L was completely resected laparo-

scopically using a SAND balloon catheter. Owing to the large diameter of MCN-L, open hepatectomy has been performed in most cases for preventing unexpected intraoperative cystic rupture.^{5,6)}

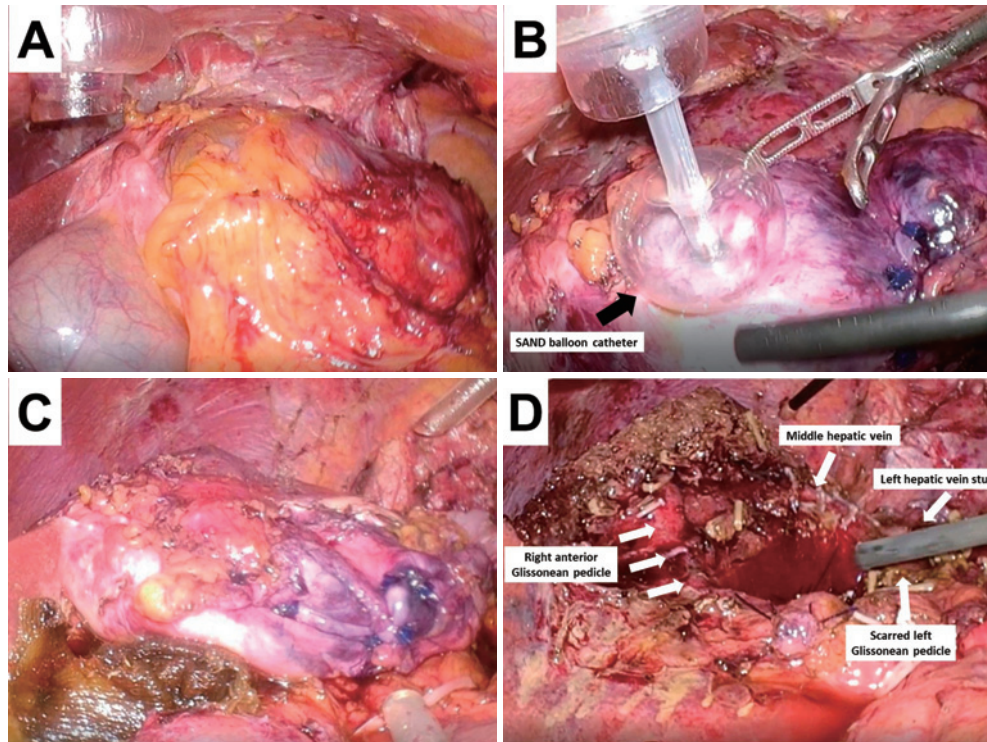


Fig. 2 Intraoperative findings

A: Before the aspiration of the cystic contents, B: Owing to the difficulty in grasping the large hepatic cystic lesion with forceps, the cystic contents were aspirated at four locations using a SAND balloon catheter. C: After the aspiration of the cystic contents, D: laparoscopic surgery can be performed completely by sequentially dissecting the Glissonean vessels toward the cyst wall.

Owing to the increasing use of LLR in recent years, however, there have been only two English reports on the use of pure LLR.^{7,8)} Obara et al.⁷⁾ performed LLR on an MCN-L with a cyst diameter of 42 mm without aspirating the cystic contents. Smerieri et al.⁸⁾ reported an MCN-L case that underwent laparoscopic left hepatectomy. Although the cyst diameter was not described, it appeared to exceed 10 cm in the images provided in the online supplementary materials. No mention was made of aspirating the cystic contents, and the leakage of cystic contents could not be ruled out because of cyst shrinking during surgery according to the online supplementary materials.

LLR for MCN-L, which is a low-grade malignant tumor, is expected to be a beneficial surgical procedure. For large MCN-L, however, there is a concern that the cystic contents may leak intraoperatively, which may lead to disseminated postoperative recurrence. Therefore, we first considered performing extended left hepatectomy or left trisegmentectomy of the liver, including caudate lobectomy by open surgery, because the large MCN-L occupied the left lobe of the liver and was in close contact with the

hepatic hilum and right anterior Glissonean pedicle. However, on the basis of the following information, we reconsidered the surgical procedure. 1) In our case, laparoscopic hepatic cystic fenestrations had been performed twice, and the pathological results were diagnosed as MCN-L with low-grade dysplasia. No intracystic mass was found in preoperative imaging, and the patient was preoperatively diagnosed as not having malignant MCN-L. 2) Because MCN-L is a rare disease, sufficient case accumulation and examination have not been conducted. The analysis with the largest number of cases was reported by Zen et al.,¹⁾ who summarized 54 cases of MCN-L. The report suggested that MCN-L is a lower-grade malignancy compared to pancreatic MCN. Fifty-three of the 54 cases (98%) were reported to have low- to moderate-grade dysplasia. 3) Most MCN-L surgical cases were reported by Lee et al.²⁾ Of the 19 MCN-L cases, radical resection was performed in 11 cases (57.9%) and liver-preserving surgery was performed in 8 cases (42.1%). No recurrence was observed in the radical resection group, but it was observed in 5 out of 8 cases in the liver-preserving surgery group. Therefore,

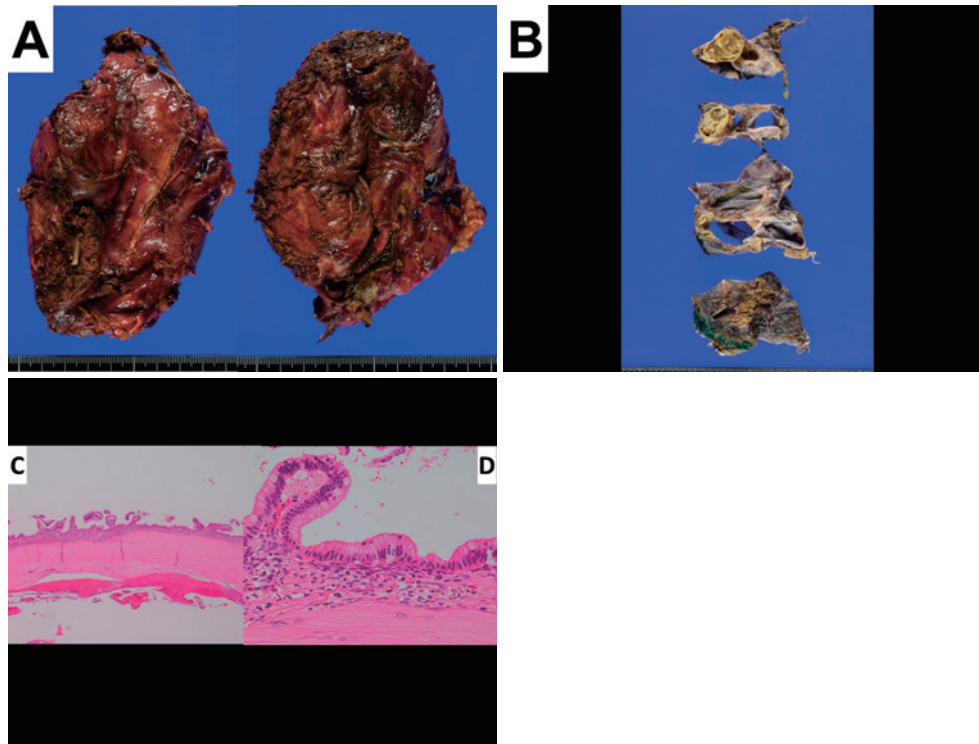


Fig. 3 Pathological findings

A, B: The resected specimen demonstrated a $13 \times 8 \times 4.5$ -cm multiloculated cyst. C: Histological examination revealed the presence of low papillary growth in certain areas ($\times 40$). D: Given the existence of ovarian-like stroma below the cystic epithelium, the patient was diagnosed with mucinous cystadenoma of the liver with low-grade dysplasia ($\times 400$).

radical resection is desirable for MCN-L. Regarding surgical procedure, the radical resection group included tumor enucleation and anatomical hepatectomy, and the liver-preserving surgery group included cystic fenestration and partial cyst resection. That is, if the entire cyst wall can be removed, extended surgery, including resection of the main vessels, will be unnecessary. Considering the three points explained above, we informed the patient multiple times regarding the surgical procedure. As a result, extended left hepatectomy or left trisegmentectomy with caudate lobectomy was over-surgery. Because the patient strongly desired laparoscopic surgery, we decided to perform the laparoscopic entire left hepatic cystectomy.

Laparoscopic surgery for large cystic tumors may result in the leakage of cystic contents due to unexpected cyst rupture during surgery,³⁾ and it is difficult to sufficiently aspirate the cystic contents during the surgery. Therefore, referring to the report by Nakamura et al.,⁴⁾ using a SAND balloon catheter for the large pancreatic MCN, we aspirated the cystic contents in advance using the SAND balloon catheter in our case.

Laparoscopic surgery using a SAND balloon catheter for mucinous cystic tumors was approved by our institutional review board, and the patient was given multiple explanations regarding the surgery. The possibility of disseminated recurrence due to mucus leakage was also explained. Finally, the patient agreed to undergo LLR using a SAND balloon catheter and provided written consent.

In our case, we applied a SAND balloon catheter used for ovarian cystic neoplasms to aspirate the cystic contents.⁹⁾ The SAND balloon catheter is a disposable, single-use, sterile device that consists of two balloons and a needle for puncturing the cyst. After puncturing the cyst, the first balloon to inflate is the distal balloon, followed by the proximal balloon.⁹⁾ The SAND balloon catheter can prevent the leakage of cystic contents by sandwiching the cyst wall with the balloon. After aspirating of the cystic contents, the cyst wall can be managed. Although the leakage of cell-level cystic content cannot be denied, no leakage of cystic contents was noted under laparoscopic view during puncture, aspiration, and suture of the puncture site. We believe that this procedure can be used for ex-

tremely particular cases such as this one.

In conclusion, for large MCN-L, LLR using a SAND balloon catheter may prevent the leakage of cystic contents and may be considered an effective device for facilitating LLR.

Author's contribution: All authors were involved in the management of the patient and in the conception of the manuscript. K.A. drafted the case report. All authors read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Ethics statement: This SAND balloon catheter is covered by insurance for ovarian cystic neoplasms and not covered by MCN-L. Therefore, off-label use of SAND balloon catheter was approved by the ethics committee and off-label use review committee of Toho University Ohashi Medical Center (H20051 and 2020-03).

Written informed consent was obtained from the patient for this procedure before surgery. In addition, the patient provided consent for the publication of this case report.

Conflicts of interest: None declared.

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