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Hepatic Resection for Cystic Lesions of the Liver

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Objective.
The purpose of this study was to report the authors' experience with hepatic resection for cystic lesions of the liver.

Summary Background Data.
Past experience with aspiration, sclerosing therapy, internal drainage, fenestration, and marsupialization are of limited value. Hepatic resection has evolved into a safe operation over the last two decades.

Methods.
A retrospective study of 44 patients with various cystic lesions of the liver (polycystic disease, 2; solitary or multiple congenital cysts, 19; biliary cystadenoma, 6; cystadenocarcinoma, 3; squamous cell carcinoma, 3; Caroli's disease, 5; and hydatid cyst, 6) was performed.

Results.
After 7 trisegmentectomies, 24 lobectomies, 6 left lateral segmentectomies, and 7 nonanatomical hepatic resections, only 1 operative death occurred in a Jehovah's Witness. Symptomatic relief was complete and permanent in all of the patients with benign congenital or parasitic hepatic cysts, except for the two patients with polycystic disease of the liver. One of the 3 patients with adenocarcinoma and 3 patients with squamous cell carcinoma of the cyst wall died of tumor recurrence between 3 and 14 months after hepatic resection.

Conclusions.
Hepatic resection is safe and effective for cystic lesions of the liver. Symptomatic relief is complete and permanent after hepatic resection, except in cases of diffuse polycystic disease of the liver. Liver transplantation should be considered for diffuse polycystic disease of the liver when the symptoms are extremely severe.

When the mortality and morbidity of hepatic resection were unacceptably high, cystic lesions of the liver were treated conservatively by repeated needle aspirations, sclerosing therapy, fenestration, marsupialization, or internal drainage.[1–5] However, hepatic resection has become a safe and effective procedure in the last two decades not only for the malignant, but also for the benign hepatic lesions.[6–8] We report our experience with hepatic resections for cystic lesions of the liver over nearly the last three decades.

MATERIALS AND METHODS

Pathology, Sex, and Age

During the 27-year period from 1964 to 1991, 740 patients with various hepatic lesions were treated by he-

Table 1. DIAGNOSIS OF THE CYSTIC LESIONS OF THE LIVER

	No. of Patients	Sex (M/F)	Mean Age in Years (Range)
Congenital cyst			
Polycystic disease	2	0/2	42.5 (36–49)
Solitary or multiple	19	3/16	55.1 (28–84)
Neoplastic cyst			
Biliary cystadenoma	6	0/6	40.8 (23–77)
Adenocarcinoma	3	0/3	55.6 (43–64)
Squamous cell carcinoma	3	2/1	50 (29–62)
Caroli's disease	5	2/3	44.6 (21–65)
Parasitic cyst	6	4/2	43 (31–71)
Total	44	11/33	47.4 (21–84)

patic resection at the University Health Sciences Center of Colorado (1964 to 1980) and the University of Pittsburgh Medical Center (1981 to 1991). Forty-four (6%) of the 740 patients had cystic lesions of the liver. There were 11 men and 33 women whose ages ranged from 21 to 84 years (mean age, 47 years). Pathologic diagnoses of the 44 patients with cystic lesions of the liver are summarized in Table 1.

Operative Procedures

For practical purposes, the types of hepatic resections were categorized as follows: right and left trisegmentectomy, right and left hepatic lobectomy, left lateral segmentectomy, and nonanatomical local resection. Extended right and left lobectomy were classified as right and left lobectomy in this report. The details of our techniques of hepatic resections were reported earlier.[9–11] The types of hepatic resection that were used to excise the cystic lesions of the liver are summarized in Table 2. Only 7 of the 44 cystic lesions were removed by nonanatomical, local resections.

Indications for Excisional Therapy (Symptoms)

The symptoms that lead to surgical excision of cystic lesions are summarized in Table 3. Severe pain caused by massive cysts was the most common reason for excision, and this was present in 28 (64%) of the 44 patients. The pain was occasionally so severe that the two patients with polycystic disease of the liver became dependent on narcotics and one suffered from rib-fracture due to expanding cysts. Abdominal fullness or tightness occurred in 14 of the 44 patients (31%) and was often associated with severe pain, anorexia/nausea, dysuria, dyspnea, and/or weight loss. The conditions of two of the patients were maintained by total parenteral nutrition before excision of the cysts.

Obstructive jaundice was the main reason for excision of cystic lesions in 12 (27%) patients, 9 of whom had clinical signs and symptoms of cholangitis. Hemorrhage in the cyst occurred in two patients. One patient with a solitary congenital cyst of the liver became suddenly anemic with abdominal pain, and another patient with cystadenocarcinoma of the liver experienced hemobilia (Figs. 1 and 2). One patient with Caroli's disease associated with choledochal cyst experienced repeated episodes of pancreatitis.

Seventeen (26%) of the 44 patients had received unsuccessful interventional or surgical treatments for their symptomatic cystic lesions of the liver (Table 4). Ten patients had had their cysts percutaneously aspirated with or without sclerosing therapy on multiple occasions to relieve their symptoms. Two patients had marsupialization and/or fenestration and three patients had internal drainage of the cyst without long-term relief. Three patients with Caroli's disease had received choledochojejunostomy without excision of the intrahepatic cystic dilation of the bile duct. One patient with a hydatid cyst had been treated with cystectomy, but the hydatid cyst recurred with symptoms.

Only 2 patients among our series of 44 patients had minimum symptoms or complaints related to the cystic lesions. Both patients had been observed for years because of minor discomforts. One of them had a sudden increase in the size, and a suspicious radiologic change in the cyst wall for a neoplasm developed in the other. The latter patient was shown to have cystadenoma at surgery.

RESULTS

Operative Mortality and Morbidity

There was only 1 operative death (a death within 1 month of surgery) among 44 hepatic resections (Table 2). This unfortunate woman was a Jehovah's Witness who had been suffering from severe right upper abdominal and back pain and repeated febrile episodes. Multiple needle aspirations had been performed for temporary symptomatic relief over several years. She died of hemorrhage during mobilization of a large, infected cyst from the densely adherent diaphragm, inferior vena cava, and right hepatic vein while right hepatic lobectomy was being performed.

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Table 2. TYPES OF HEPATIC RESECTION AND OPERATIVE MORTALITY

	Right Trisegmentectomy	Left Trisegmentectomy	Right Lobectomy	Left Lobectomy	Left Lateral Segmentectomy	Nonanatomical Resection	Death
Polycystic	1	—	1	—	—	—	1
Solitary/multiple	2	—	10 (1)*	3	2	2	1
Neoplastic	2	1	2	2	2	3	—
Caroli/choledocal	—	—	1	2	1	1	—
Parasitic	1	—	1	2	1	1	—
Total	6	1	15	9	6	7	1

* Operative mortality (death within 1 month) in only one patient.

Nine major complications developed in eight patients. Three cases of subphrenic abscess occurred: 12 days after right lobectomy in 1 case and 20 and 60 days after right trisegmentectomy in 2 cases. One patient had subarachnoid hemorrhage on the fourth postoperative day after nonanatomical resection for a congenital solitary cyst. Two patients were re-explored for bleeding: one immediately after right trisegmentectomy and another 1 week after left lateral segmentectomy. One patient with Caroli's disease had pulmonary embolism 10 days after left lobectomy. All of these patients recovered from these complications without sequelae. Obstructive jaundice developed 2 months after right trisegmentectomy in a patient with neoplastic cysts. Multiple episodes of cholangitis occurred 2 months after left trisegmentectomy in one case of squamous cell carcinoma due to tumor recurrence.

Symptomatic Relief and Long-term Follow-up

The two patients with severe polycystic disease of the liver had marked symptomatic relief for 13 years, but the cystic enlargement of the remaining liver eventually grew back almost to the original size. One of the patients received successful orthotopic liver transplantation for progressive disabling symptoms 8 years after hepatic resection. Another patient remains less symptomatic than before hepatic resection, but her condition is now maintained with hemodialysis for her failed polycystic kidneys.

However, 16 of the 19 patients with congenital solitary or multiple cysts are free of symptoms 1 to 9 years after hepatic resection. Of the remaining three patients, one patient died intraoperatively as previously described (a Jehovah's Witness), another died 16 months later of extrahepatic bile duct cancer that was found incidentally and that was resected at the time of surgery for the congenital cyst, and the other was lost to follow-up 1 year after surgery when she was completely asymptomatic.

All 6 patients with biliary cyst adenoma are alive and free of symptoms at 15, 24, 26, 30, 31, and 75 months after surgery. One of the three patients with adenocarcinoma of the cyst wall died with metastatic disease 3 months after surgery. The other two are alive: one free of disease at 73 months and another with tumor recurrence

Table 3. SIGNIFICANT SYMPTOMS CAUSED BY CYSTIC LESIONS OF THE LIVER

Symptoms	Polycystic Disease (n = 2)	Solitary/Multiple Cysts (n = 19)	Neoplastic Cysts (n = 12)	Caroli's Disease (n = 5)	Parasitic Cysts (n = 6)	Total (n = 44)
Abdominal pain	2	13	9	—	4	28
Abdominal mass/fullness	2	4	7	1	—	14
Dyspnea	1	—	—	—	—	1
Anorexia/nausea	2	1	2	—	—	5
Weight loss	2	1	2	—	—	5
Jaundice/cholangitis	—	3	2	3	4	12
Hemorrhage	—	1	1	—	—	2
Pancreatitis	—	—	—	1	—	1
Dysuria	1	—	—	—	—	1
Nonspecific/minor	—	1	—	—	—	1

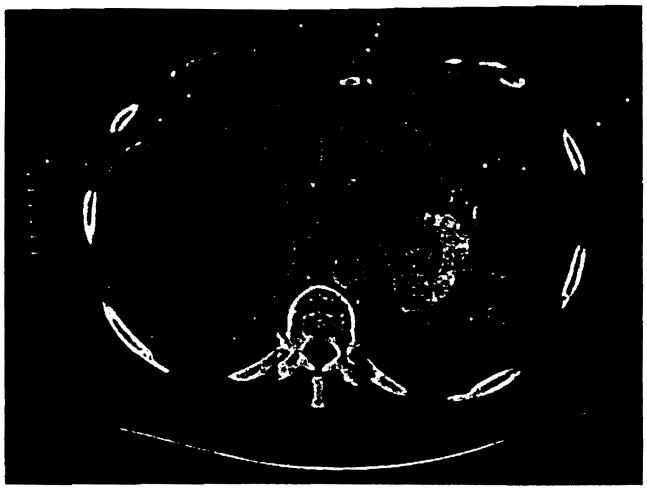

Figure 1. Computed axial tomography scan showing internal hemorrhage and rupture of the congenital cyst.

26 months after right lobectomy. The three patients with squamous cell carcinoma arising in the cyst wall died of recurrent tumor at 4, 8, and 14 months after hepatic resection.

All five patients with Caroli's disease are alive and free of symptoms at 12, 42, 69, 78, and 84 months after surgery.

One of the six patients with hydatid cyst died of pancreatic cancer 13 months after local excision of the cyst. The other five patients are asymptomatic and free of cyst recurrence at 18, 30, 62, 78, and 180 months after hepatic resection.

DISCUSSION

The safety and effectiveness of hepatic resection in treating cystic lesions of the liver has been proven in this report. One operative death occurred in a Jehovah's Witness, and only 9 major complications developed in 8 patients in this series of 44 patients. The relief from significant symptoms was complete and permanent, except in two patients with polycystic disease.

Aspiration, sclerotherapy, internal drainage, marsupialization, and fenestration[1-5] have all been recommended for congenital hepatic cysts. These therapies can no longer be justifiable treatments of first choice for single or localized multiple cysts because the resection can be performed so safely and effectively.[6-8]

The diagnosis of neoplastic cyst could not be made preoperatively in any of the 12 patients. Four of these patients had previous aspirations and two had internal drainage through jejunal Roux limbs of the cyst elsewhere. Pain, fever, and intermittent jaundice recurred after internal drainage of the cysts. By the time we performed resection, squamous cell carcinoma of the cyst wall had spread regionally in the three patients. They died of widespread cancer between 6 and 14 months. Al-

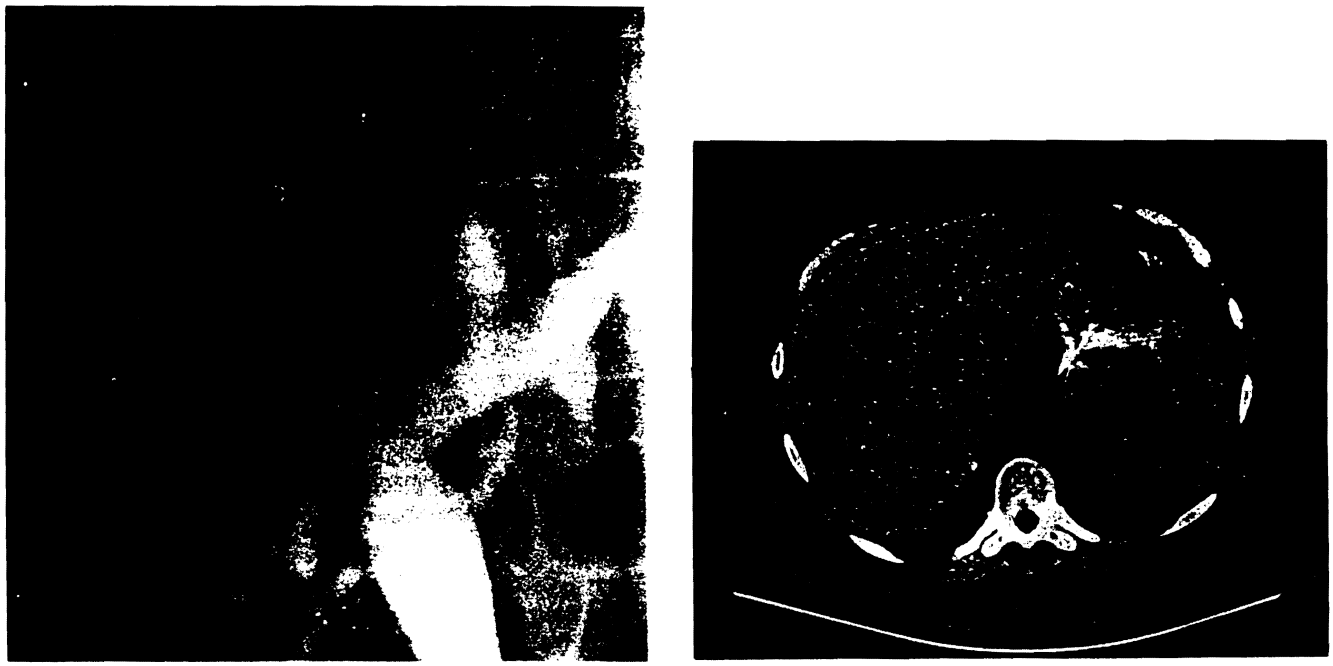

Figure 2. (Left) An endoscopic retrograde cholangiogram demonstrating communication of the cyst with the bile duct in a patient with cystadenocarcinoma who experienced hemobilia. (Right) Computed axial tomography scan of the same patient demonstrating contrast material and blood clot within the cyst.

Table 4. PREVIOUS TREATMENTS

	Polycystic Disease	Solitary/ Multiple Cysts	Neoplastic Cysts	Caroli's Disease	Parasitic Cysts	Total
Aspiration/percutaneous drainage	3	3	4	—	—	10
Marsupialization	1	—	—	—	—	1
Fenestration	1	—	—	—	—	1
Internal drainage	—	—	2	1	—	3
Excision	—	—	—	—	1	1
Sclerosing therapy	—	—	—	—	1	1
Total	5	3	6	1	2	17

though the incidence of malignancy developing in the cyst wall is low,[12-16] the survival could be improved if the symptomatic solitary or multiple congenital hepatic cysts are primarily treated by hepatic resection.

Marsupialization, fenestration, and resection of most of the cysts should be applied only for exceptional cases of polycystic liver disease.[17,18] In our two patients with widespread polycystic disease, both had marked immediate symptomatic relief, but the cyst eventually grew back in the remaining portion of the liver almost to the original size. One of them required orthotopic liver transplantation 8 years after resection. Her transplantation was extremely difficult because of dense adhesions that had been created by previous operation. We have successfully treated several other patients with severe polycystic disease involving both the liver and the kidneys by combined liver and kidney transplantation. The quality of life of these transplanted patients is much better than that of patients after partial hepatic resection.[19] Confronted with extreme complications of polycystic disease today, we would recommend organ replacement as the first, not the last, step of therapy. Otherwise, the polycystic disease of the liver should not be carelessly treated surgically.

References

1. Sanfelippo PM, Beahrs OH, Weiland LH. Cystic disease of the liver. Ann Surg 1974; 179:922–925.
2. Andersson R, Jeppsson B, Lunderquist A, Bengmark S. Alcohol sclerotherapy of non-parasitic cysts of the liver. Br J Surg 1989; 76: 254–255.
3. Saini S, Mueller PR, Ferrucci JT Jr, et al.. Percutaneous aspiration of hepatic cysts does not provide definitive therapy. AJR Am J Roentgenol 1983; 141:559–560.
4. Lai ECS, Wong J. Symptomatic nonparasitic cysts of the liver. World J Surg 1990; 14:452–456.
5. Armitage NC, Blumgarten LH. Partial resection and fenestration in the treatment of polycystic liver disease. Br J Surg 1984; 71:242–244.
6. Iwatsuki S, Starzl TE. Personal experience with 411 hepatic resections. Ann Surg 1988; 208:421–434.
7. Iwatsuki S, Sheahan DG, Starzl TE. The changing face of hepatic resection. Curr Probl Surg 1989; 26:289–379.
8. Iwatsuki S, Todo S, Starzl TE. Excisional therapy for benign hepatic lesions. Surg Gynecol Obstet 1990; 171:240–246.
9. Starzl TE, Bell RH, Beart RW, Putnam CW. Hepatic trisegmentectomy and other liver resections. Surg Gynecol Obstet 1975; 141: 429–437.
10. Starzl TE, Koep LJ, Weil R, et al. Right trisegmentectomy for hepatic neoplasms. Surg Gynecol Obstet 1980; 150:208–213.
11. Starzl TE, Iwatsuki S, Shaw BW Jr, et al. Left hepatic trisegmentectomy. Surg Gynecol Obstet 1982; 155:21–27.
12. Bloustein PA, Silverberg SG. Squamous cell carcinoma originating in an hepatic cyst. Cancer 1976; 38:2002–2005.
13. Richmond HG. Carcinoma arising in congenital cysts of the liver. J Pathol Bacteriol 1956; 72:681–683.
14. Lynch MJ, McLeod MK, Weatherbee L, et al. Squamous cell cancer of the liver arising from a solitary benign nonparasitic hepatic cyst. Am J Gastroenterol 1988; 83:426–431.
15. Ishak KG, Willis GW, Cummins SD, Bullock AA. Biliary cystadenoma and cystadenocarcinoma. Cancer 1977; 39:322–338.
16. Bloustein PA. Association of carcinoma with congenital cystic conditions of the liver and bile ducts. Am J Gastroenterol 1977; 67: 40–46.
17. Newman KD, Torres VE, Rakela J, Nagorney DM. Treatment of highly symptomatic polycystic liver disease. Ann Surg 1990; 212: 30–37.
18. Van Erpecum KJ, Janssens AR, Terpstra JL, Tham RTO. Highly symptomatic adult polycystic disease of the liver. J Hepatol 1987; 5:109–117.
19. Starzl TE, Reyes J, Tzakis A, et al. Liver transplantation for polycystic liver disease. Arch Surg 1990; 125:575–577.