J Korean Radiol Soc 1998;39:773-777

Intrahepatic Cholangiocarcinoma Presenting as Liver Abscess : Report of Two Cases[1]

Kwon-Hyoung Kim, M.D., On-Koo Cho, M.D., Yong-Soo Kim, M.D.,
Hyun-Chul Rhim, M.D., Byung-Hee Koh, M.D..

Intrahepatic cholangiocarcinoma is the second most common primary malignant hepatic neoplasm. We describe two cases of intrahepatic cholangiocarcinoma which initially presented as liver abscess both clinically and radiologically. Mucin-hypersecretion from the tumor cells and extensive necrosis or secondary bacterial infection was responsible for the radiologic appearance of a liver abscess.

Index words : Bile ducts, CT
Bile ducts, US
Bile ducts, neoplasms
Liver, abscess

Intrahepatic cholangiocarcinoma usually appears to be a solid mass which shows low attenuation on pre-enhanced CT and homogeneous hyperechogenecity or mixed echogenecity on sonography (1, 2). Common clinical presentations are abdominal pain, anorexia and weight loss. Initial presentation as a liver abscess is extremely rare clinically and radiologically. To our knowledge, only two cases of intrahepatic cholangiocarcinoma mimicking liver abscess radiologically have been reported (3, 4). We describe two cases of intrahepatic cholangiocarcinoma which initially presented as liver abscess both clinically and radiologically.

Case Report

Case 1

A 73 year-old male presented with abdominal discomfort and fever of up to 39.2°C, which lasted 10 days. Laboratory data showed leukocytosis of 18, 600/mm^3. Initial contrast enhanced CT showed a large cystic mass of 12 × 9 cm in the right lobe of the liver, with a double target sign which was highly suggestive of liver abscess (Fig. 1A). About 1500 cc of chocolate colored and odorless turbid fluid was aspirated from the cystic mass. Enterococcus was positive on Gram stain and culture; cytologic examination of the aspirated fluid failed to reveal any malignant cells. Antibiotics were infused intravenously with subsequent symptomatic improvement and the patient was discharged. He was, however, admitted again with the same symptoms one year later. At that time, abdominal US revealed a 5 × 6 cm cystic mass with an echogenic component in the right lobe of the liver (Fig. 1B). Contrast enhanced CT revealed a cystic mass with increased soft tissue density in the medial portion of the cavity and dilatation of adjacent intrahepatic bile duct (Fig. 1C). ERCP and abscessogram through a pig tail catheter showed communication between the cystic cavity and intrahepatic bile ducts, as well as multiple filling defects in the intrahepatic bile duct and common bile duct (Fig. 1D). A Right lobectomy was performed. Gross specimen showed a large cystic mass and papillary tumor growth in the wall of the cystic mass (Fig. 1E), and communication between the cystic mass and dilated intrahepatic bile duct was confirmed. Microscopic examination revealed marked dilatation of intrahepatic bile ducts and papillary configuration of adenocarcinoma, with columnar epithelial cells around thin fibrovascular cores (Fig. 1F). The final diagnosis was intrahepatic papillary cholangiocarcinoma.

[1]Department of Diagnostic Radiology, College of Medicine, Hanyang University
Received June 25, 1998 ; Accepted August 3, 1998
Address reprint requests to : On-Koo Cho, M.D., Department of Diagnostic Radiology, College of Medicine, Hanyang University # 17, Haengdang-Dong, Sungdong-Gu Seoul 133-792, Korea Tel. 82-2-290-9164 Fax. 82-2-293-2111

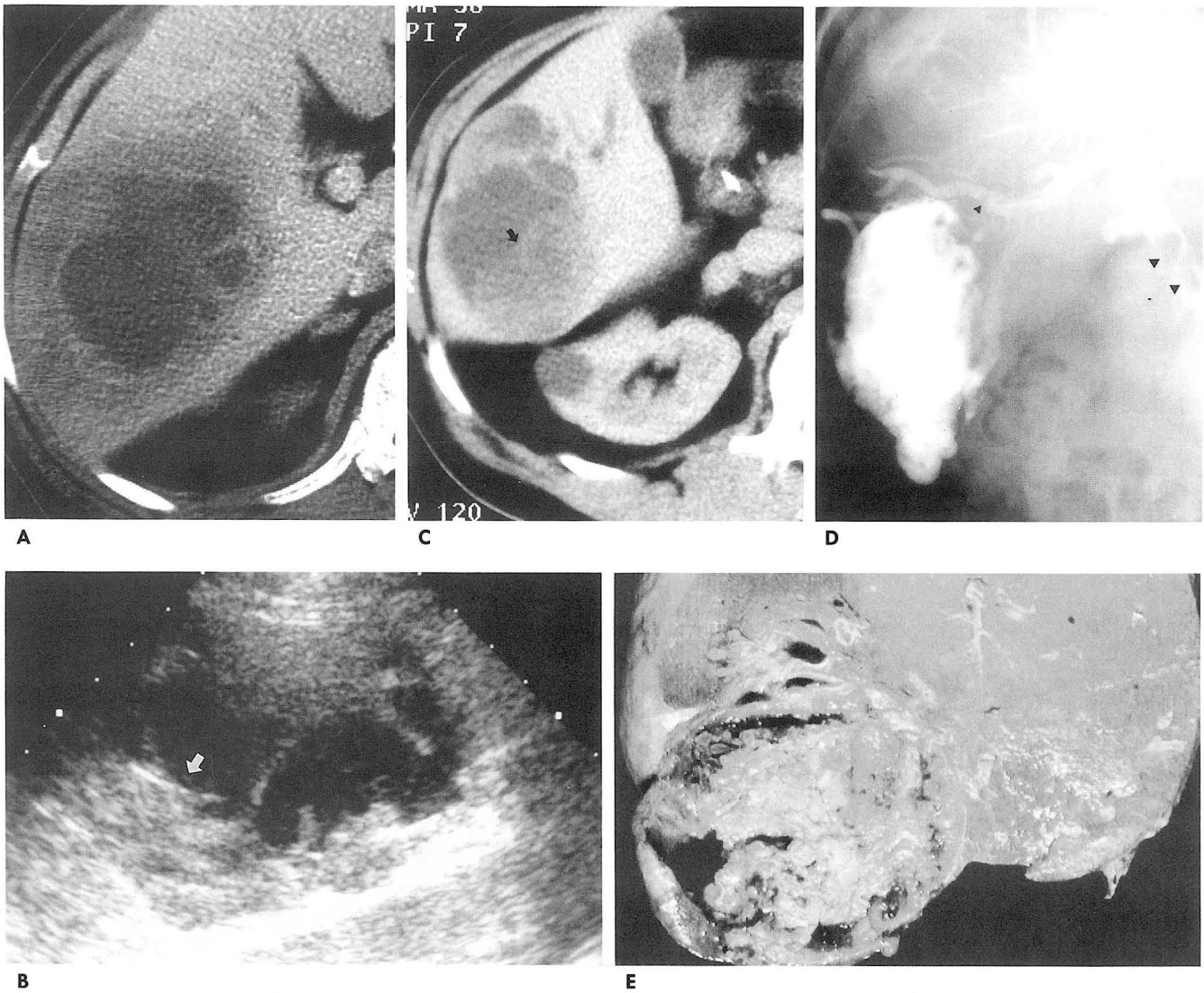

Fig. 1. A 73-year-old man with abdominal discomfort and fever (Case 1)

A. Contrast enhanced CT scan shows a large cystic mass in the right lobe of liver with double target sign characteristic of liver abscess.

B. Ultrasonogram obtained one year later shows irregular shaped cystic mass with eccentric echogenic component (arrow).

C. Contrast enhanced CT scan reveals a cystic mass with ill-defined area of slightly increased soft tissue density (arrow) in the medial wall of the cyst.

D. Abscessogram reveals multiple amorphous filling defects (arrowheads) in the CBD and IHD and communication between the cystic mass and adjacent IHD.

E. Gross specimen shows marked cystic dilatation of the IHD and papillary tumor growth within it.

F. Mircroscopic examination reveals papillary configuration of tumor growth with columnar epithelial cells around thin fibrovascular core (H & E stain, × 100).

Case 2

A 65 year-old female was admitted due to fever of up to 38°C and right flank pain of two months' duration. Initial ultrasonography showed a lobulating hypoechoic mass in the right lobe of the liver (Fig. 2A). Contrast enhanced CT scanning revealed a 7 × 5 cm, irregular shaped cystic mass with thick enhancing peripheral wall (Fig. 2B). The inner margin of the cystic mass was irregular and some papillary projection was noted. Yellowish turbid fluid was aspirated from the cystic mass. Gram stain and culture of the aspirate showed no growth or staining of pathogenic organisms and cytologic examination showed no malignant cells. Laboratory data included leukocyte count of 8,000/mm^3, carcinoembryonic antigen of 3.9 IU/ml, and alpha-fetoproten of 1.7 IU/ml. Initial diagnostic impression was liver abscess. Follow-up sonography and CT failed to show a decrease in the size of the cystic mass after percutaneous drainage. An abscessogram taken through the drainage catheter showed opacification of the cystic cavity with irregular and papillary margin and communication between the cystic cavity and the intrahepatic bile duct (Fig. 2C). A right lobectomy was performed. Gross and microscopic examination of the specimen revealed that the cystic mass was an intrahepatic mucoepidermoid carcinoma with exten-

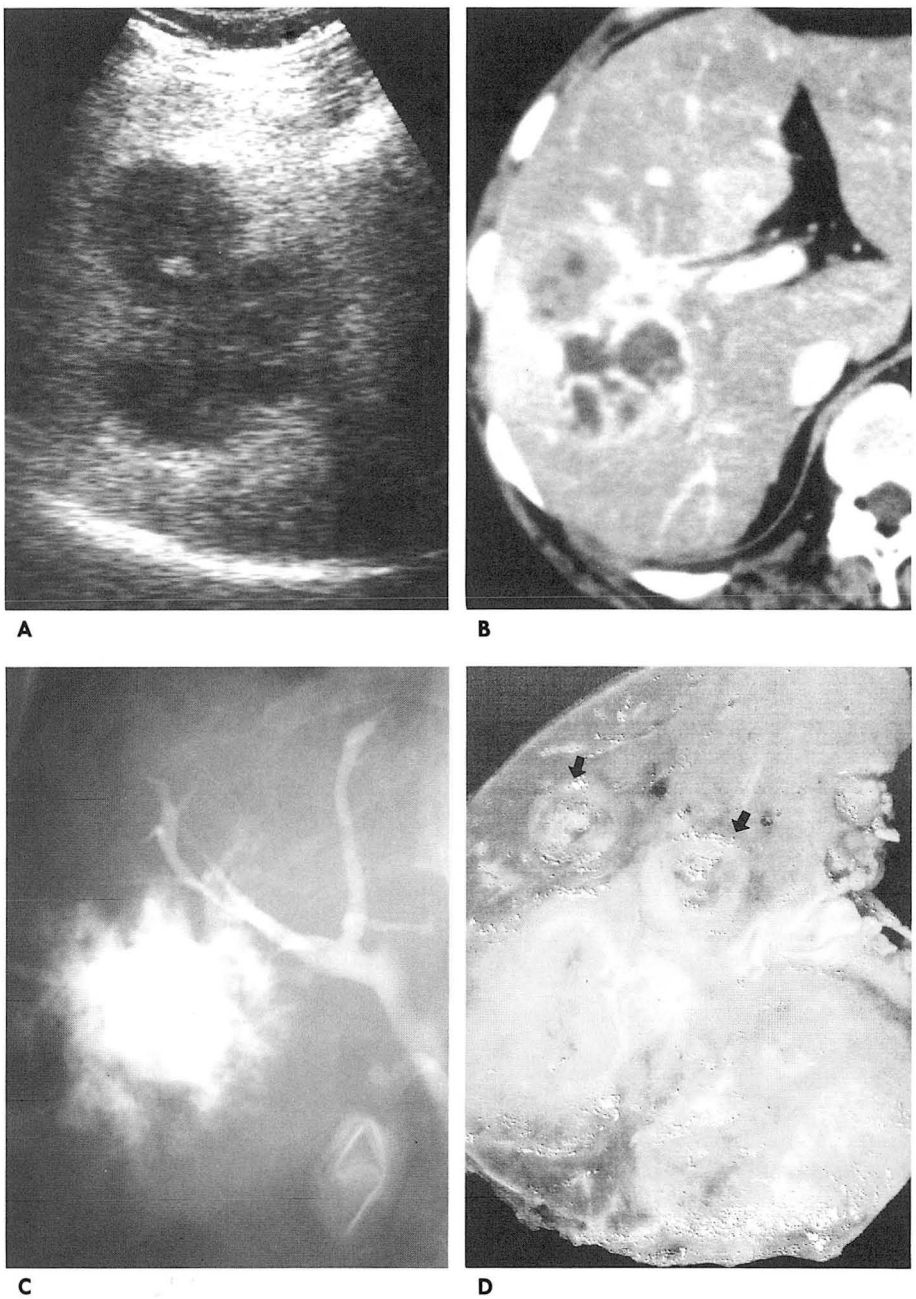

Fig. 2. A 65-year-old woman with fever and right flank pain (Case 2)
A. Ultrasonogram of the right lobe of liver shows lobulated hypoechoic mass with slightly echogenic internal septum-like component.
B. Contrast enhanced CT scan reveals multiloculated cystic mass with thick enhancing peripheral wall and internal septum.
C. Abscessogram through drainage catherter shows opacification of the cystic cavity with papillary border and contrast filling of IHD and CBD indicating the presence of communication with IHD.
D. Gross examination of the surgical specimen reveals a large mass of lobulating contour with extensive central necrosis. Multiple satellite nodules (arrows) are seen around the main mass.

sive central necrosis (Fig. 2D). The solid component of the mass was composed of epidermoid, mucin-producing cells and ductal components. Mucoepidermoid carcinoma was confirmed.

Discussion

Intrahepatic cholangiocarcinoma is thought to originate from the epithelial cells lining the intrahepatic bile duct. On the basis of location, it is subdivided into central and peripheral types ; the latter origintates distal to second-order branches of the bile duct ; and the former from the main or first branches of bile ducts. CT findings of peripheral cholangiocarcinomas have been described in several reviews (1, 5). The most common CT finding is a homogeneous low attenuation mass with irregular demarcation as seen on precontrast scans and delayed tumoral enhancement (1, 6). Homogeneously hyperechoic or mixed echogenic mass is the usual sonographic finding of peripheral cholangiocarcinoma ; the gross and microscopic appearances of such a mass have been shown to correlate closely with CT and sonographic findings. Grossly, peripheral cholangiocarcinoma usually appears to be a large, white, firm solid tumor ; microscopically it is known to contain abundant fibrous stroma and collagen fibers, which play an important role in delayed contrast enhancement on CT (6).

The CT and sonographic findings of our two cases did not show the usual pattern of peripheral cholangiocarcinomas, previously described. Initial radiological and clinical presentation of this carcinoma as liver abscess is extremely rare ; in both cases, cystic degeneration of the tumor and secondary bacterial infection were responsible for the CT appearance mimicking liver abscess. In our cases, the CT and sonographic findings correlated closely with pathologic findings.

In case 1, the cystic mass was severely dilated intrahepatic bile duct, with papillary growth of tumor from the wall of the cyst. The final diagnosis was intrahepatic papillary cholnagiocarcinoma. About 5 % of all cholangiocarcinomas are papillary. Intrahepatic papillary cholangiocarcinomas produce variable amount of mucin, but sufficient mucin production to produce a cystic mass as in cases of biliary cystadenoma or cystadenocarcinoma is reported to be rare (7). In our case of papillary cholangiocarcinoma, however, mucin-hypersecretion from the tumor cell was responsible for the radiologic appearance of a cystic mass along with secondary bacterial infection. Evidence of mucin-hypersecretion was seen on ERCP and abscessogram as multiple filling defects in the biliary trees. Extensive central necrosis of the tumor and, in part, mucin-hypersecretion were responsible for the abscess-like appearance in the other tumor, which was finally diagnosed as mucoepidermoid carcinoma. Intrahepatic mucoepidermoid carcinoma is a rare entity and is considered to be a variant of cholangiocarcinoma ; it consists of squamous cells, mucus-producing cells and glandular cells (8—10). We found no report of this rare entity in the radiological literature. In the pathologic report describing one case of mucoepidermoid carcinoma, this appeared as a low attenuated mass which on CT showed central water-density area in the liver. Microscopically, the tumor was intimately contiguous with a preexisting cyst and gradual transition between tumor cells and benign cystic lining cells was recognized, suggesting that the mucoepidermoid carcinoma was derived from malignant transformation of a preexisting cyst (9). Another case of mucoepidermoid carcinoma of the liver was associated with multiple sero-mucinous cyst (10). Communication with intrahepatic bile ducts was not found in any case previously reported.

Percutaneous transhepatic drainage is the basis for most biliary interventional procedures. Although the incidence is low, dissemination of tumor cells along the transhepatic catheter tract is considered to be a serious complication of percutaneous transhepatic biliary drainage in patients with biliary tract obstruction due to cholangiocarcinoma or pancreatic cancer (11) and it is thus generally recommended that when curative resection is planned, PTBD should be avoided. Fortunately, in our cases, no evidence of seeding of tumor cells was seen grossly or microscopically.

Finally, we concluded that intrahepatic cholangiocarcinoma may initially present as liver abscess radiologically and clinically, although the incidence is very low. It must be included as an alternative diagnosis in patients with liver abscess which is longstanding and intractable despite medical treatment and drainage, especially in cases showing communication with intrahepatic bile duct.

References

1. Lacomis JM, Baron RL, Oliver JH, Nalesnik MA, Federle MP. Cholangiocarcinoma : delayed CT contrast enhancement patterns. *Radiology* 1997 ; 203 : 98-104
2. Wibulpolprasert B, Dhiensiri T. Peripheral cholangiocarcinoma : sonographic evaluation. *J Clin Ultrasound* 1992 ; 20 : 303-314
3. Kokubo T, Itai Y, Nagao T. Intrahepatic cholangiocarcinoma with cystic formation. *Radiat Med* 1990 ; 8 : 219-221
4. Pramoolsinsap C, Kurathong S. Cholangiocarcinoma masquerading

as liver abscess. *Southest Asian J Trop Med Public Health* 1989 ; 20 : 313-317
5. Ros PR, Buck JL, Goodman ZD, Ros AM, Olmsted WW. Intrahepatic cholangio- carcinoma : radiologic-pathologic correlation. *Radiology* 1988 ; 167 : 689-693
6. Itai Y, Ohtomo K, Kokubo T, et al. CT of hepatic masses : significance of prolonged and delayed enhancement. *AJR* 1986 ; 146 : 729-733
7. Kobubo T, Itai Y, Ohtomo K, et al. Mucin-hypersecreting intrahepatic biliary neoplasms. *Radiology* 1988 ; 168 : 609-614
8. Di Palma s, Andreola S, Audisio RA, Doci R, Lombardi L. Primary mucoepidermoid carcinoma of the liver. a case report. *Tumori* 1992 29 ; 78 : 65-68
9. Hayashi I, Tomoda H, Tanimoto M, et al. Mucoepidermoid carcinoma arising from a preexisting cyst of the liver. *J Surg Oncol* 1987 ; 36 : 122-125
10. Katsuda S, Nakanishi I, Kajikawa K, Takabatake S. Mucoepidermoid carcinoma of the liver. *Acta Pathol Jpn* 1984 ; 34 : 153-157
11. Chapman WC, Sharp KW, Weaver F, Sawyers JL. Tumor seeding from percutaneous biliary catherters. *Ann surg* 1989 ; 209 : 708-713

대한방사선의학회지 1998;39:773-777

간농양으로 나타난 간내담관암 : 2예 보고

한양대학교 의과대학 진단방사선과학교실

김권형 · 조온구 · 김용수 · 임현철 · 고병희

간내담관암은 두 번째로 흔한 악성 간종양이다. 저자들은 임상적, 방사선학적으로 간농양으로 나타난 간내담관암 2예를 보고하고자 한다. 저자들의 증례에서는 종양세포의 뮤신과다분비, 종양의 광범위한 괴사와 이차적인 세균감염등에 의하여 간농양의 방사선학적 소견을 보였다.

제42회 전문의자격시험 1차시험 출제계획서

(객관식)

영역 / 수준 / 분야	X-선				혈관중재				초음파				CT				MR				RI				복합				의학				총
	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단
호흡기	1	2		3			1	1					2	3	2	7		1		1						1		1	2			2	5	7
심맥관		1		1		1	1	2						1		1		1		1									1			1	1	4
위장관	2	2		4					1			1		1	1	2										1		1	1			1	4	4
간·담도·췌		1		1		1		1	1	1		2		1	1	2	1	1		2					1			1					3	5
비뇨·생식	1			1			1	1	1	1		2	1	1		2		2		2						1	1	2	1			1	4	5
신경						1	1	2					1	1		2	2	2	1	5					2	1		3	1	1		2	6	6
근골격		2	1	3						1		1		1		1	1	1		2						1		1	1			1	2	6
소아	1	1		2					1	1		2	1		1	2	1	1		2							1	1	1			1	5	3
유방		1		1					1			1													1			1				2	1
핵의학																					2	2		4		1	1	2	1			1	3	3
물리	1			1					1			1		1		1		1	1	2									1			1	3	2
법규																													1			1	1	
총 계	6	10	1	17		3	4	7	6	4		10	5	10	5	20	5	10	2	17	2	2		4	4	6	3	13	11	1		12	39	46

(주관식)

영역 / 수준 / 분야	X-선				혈관중재				초음파				CT				MR				RI				복합				의학				총
	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단	해결	소계	암기	판단
호흡기			1	1									1	2		3											1	1	1			1	2	2
심맥관						1	1	2																										1
위장관	1			1										1		1										1		1	1			1	2	2
간·담도·췌										1		1	1			1		1		1						1		1					1	3
비뇨·생식	1			1						1		1			1	1	1			1						1		1					2	2
신경						1		1						1		1	1	1	1	3									1			1	2	3
근골격		1		1														1		1							1	1	1			1	1	2
소아		1		1						1		1						1		1									1			1	1	3
유방		1		1																														1
핵의학																						1		1	1			1					1	1
물리	1			1													1			1													2	
법규																																		
총계	3	3	1	7		2	1	3		3		3	2	4	1	7	3	4	1	8		1		1	1	3	2	6	5			5	14	20