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CASE REPORT

Sarcomatoid intrahepatic cholangiocarcinoma mimicking liver abscess: A case report

Yan Wang, Jia-Lei Ming, Xing-Yu Ren, Lu Qiu, Li-Juan Zhou, Shu-Dong Yang, Xiang-Ming Fang

ORCID number: Yan Wang (0000-0002-5151-922X); Jia-Lei Ming (0000-0002-3536-4694); Xing-Yu Ren (0000-0001-6122-6837); Lu Qiu (0000-0001-5510-9589); Li-Juan Zhou (0000-0001-5384-2971); Shu-Dong Yang (0000-0001-8894-9314); Xiang-Ming Fang (0000-0003-2961-0809).

Author contributions: Wang Y designed the work, analyzed and interpreted the data and prepared the manuscript; Ming JL, Ren XY, Qiu L and Zhou LJ collected clinical data and edited the manuscript; Yang SD analyzed the pathological data; Fang XM was responsible for revision of the manuscript. All authors read and approved the final manuscript.

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Yan Wang, Jia-Lei Ming, Xing-Yu Ren, Lu Qiu, Li-Juan Zhou, Xiang-Ming Fang, Department of Radiology, The Affiliated Wuxi People's Hospital of Nanjing Medical University, Wuxi 214023, Jiangsu Province, China

Shu-Dong Yang, Department of Pathology, The Affiliated Wuxi People's Hospital of Nanjing Medical University, Wuxi 214023, Jiangsu Province, China

Corresponding author: Xiang-Ming Fang, MD, PhD, Chief Doctor, Instructor, Professor, Department of Radiology, The Affiliated Wuxi People's Hospital of Nanjing Medical University, No. 299 Qingyang Road, Wuxi 214023, Jiangsu Province, China.
xiangming_fang@njmu.edu.cn

Abstract

BACKGROUND

Sarcomatoid intrahepatic cholangiocarcinoma (SICC) is an extremely rare and highly invasive malignant tumor of the liver. To our knowledge, the imaging findings of sarcomatous cholangiocarcinoma have been rarely reported; and radiological features of this tumor mimicking liver abscess have not yet been reported.

CASE SUMMARY

We present a case of SICC mimicking liver abscess. The patient, a 43-year-old male, complained of repeated upper right abdominal discomfort and intermittent distension over a period of one month. Radiology examination revealed a huge focal lesion in the right liver. The lesion was hypointense on computed tomography with honeycomb enhancement surrounded by enhanced peripheral areas. It showed a hypo-signal on non-contrast T1-weighted images and a hyper-signal on non-contrast T2-weighted images. Radiologists diagnosed the lesion as an atypical liver abscess. The patient underwent a hepatectomy. After surgery, he survived another 2.5 mo before passing away. A search of PubMed and Google revealed 43 non-repeated cases of SICC reported in 20 published studies. The following is a short review in order to improve the diagnostic and therapeutic skills in cases of SICC.

CONCLUSION

This report presents the clinical and radiological features of SICC and imaging features which showed hypovascularity and progressive enhancement. SICC can present as a multilocular cyst on radiological images and it is necessary to distinguish this lesion from an atypical abscess. Simple surgical treatment is not the best treatment option for this disease.

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Core tip: Sarcomatoid intrahepatic cholangiocarcinoma (SICC) is an extremely rare and highly invasive malignant tumor of the liver. The radiological features of this lesion have been rarely reported and it is difficult to differentiate this tumor from a liver abscess. We present the clinical, radiological characteristics and treatment of a patient with SICC followed by a summary of relevant English literature regarding this condition, including a prognosis analysis of SICC.

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INTRODUCTION

Sarcomatous cholangiocarcinoma is a rare intrahepatic malignant tumor, accounting for 4.5% of intrahepatic cholangiocarcinomas[1]. Spindle-shaped and pleomorphic cells and adenoid structures are observed in sarcomatous cholangiocarcinoma[1]. In the relevant literature, it is also known as cholangiocarcinoma with sarcomatous changes[2]. The tumor pathogenesis has not been sufficiently clarified. The prognosis of sarcomatous cholangiocarcinoma is worse than that of conventional cholangiocarcinoma[1]. Pathological diagnosis is the gold standard for diagnosing sarcomatous cholangiocarcinoma. However, a preoperative biopsy can lead to intra-tumor bleeding and dissemination[2]. It is imperative to improve the preoperative diagnosis using imaging examination. To our knowledge, the imaging findings of sarcomatous cholangiocarcinoma have rarely been reported, nor have its radiological features mimicking liver abscess. We report a patient with intrahepatic sarcomatoid cholangiocarcinoma (SICC) and discuss the imaging and clinical features of this unusual disease through a careful review of the literature.

CASE PRESENTATION

Chief complaints

The patient was a 43-year-old male suffering from repeated upper right abdominal discomfort and intermittent distension over a period of one month. There was no report of radiation of the pain to his shoulder and back.

History of present illness

There was neither a history nor symptoms of fever, yellowish eyes, weight loss, or vomiting.

History of past illness

He had a 25-year history of hepatitis B. He did not take any routine medication.

Personal and family history

His medical and family history did not reveal any history of serious or terminal illnesses or any other relevant information.

Physical examination upon admission

His temperature was 37°C, resting heart rate was 80 bpm, respiratory rate was 15 breaths/min, and blood pressure was 120/80 mmHg.

Laboratory examinations

The results of blood analysis were as follows: Red blood cells, 5.08 × 10^{12}/L [normal range: (3.5-5.5) × 10^{12}/L]; white blood cells, 7.7 × 10^{9}/L [normal range: (4-10) × 10^{12}/L], and platelet count, 149 × 10^{9}/L [normal range: (80-300) × 10^{9}/L]. The results of liver

function tests were as follows: Albumin, 39.3 g/L [normal range: (35.0-53.0) g/L]; globulin, 25.5 g/L [normal range: (17.0-33.5) g/L]; lactic dehydrogenase, 181.0 U/L [normal range: (109.0-245.0) U/L]. The results of C-reactive protein and procalcitonin were 4 mg/L [normal range: (0-8) mg/L] and 0.04 ng/mL [normal range: (0-0.5) ng/mL], respectively. Other measurements were as follows: Serum α-fetoprotein (AFP) 66.91 ng/mL (normal range < 10 ng/mL), carcinoembryonic antigen 1.02 ng/mL (normal range < 5 ng/mL), serum carbohydrate antigen (CA) 19-9 and CA 125 levels were 19.9 U/mL and 26.3 U/mL (normal range: < 35 U/mL), respectively.

Imaging examinations

An abnormal mass of hypointensity on T1-weighted images (T1WI) and hyperintensity on T2-weighted images (T2WI) was observed in the right liver lobe (Figure 1). On post-contrast T1WI, honeycomb-like continuous enhancement, with slight transient hepatic parenchymal enhancement (THPE) around it, and adjacent proximal bile duct dilatation with enhancement of the wall were observed. In the arterial phase, blood vessels could be seen entering the lesion. Initially, a hepatic abscess could not be excluded. Further computed tomography (CT) examination (Figure 2) showed that the right lobe of the liver had a patchy low-density lesion of approximately 7.0 cm × 5.6 cm with heterogeneous enhancement. Mild dilation of the intrahepatic bile duct and enlarged lymph nodes of the retroperitoneal region were observed.

The diagnosing physician considered it to be a malignant occupation; thus, surgical resection was performed. During the operation, no ascitic fluid was found in the abdominal cavity. The liver was reddish-brown and nodular hyperplasia of the liver could be seen. A mass in the right lobe of the liver was observed, which had a hard texture. In addition, no satellite lesions were found around this mass. The surgeon diagnosed it as liver cancer and performed radical hepatectomy and lymphadenectomy.

The gross cross-section examination revealed a yellowish-white mass. On microscopic examination, the tumor consisted of an adenocarcinoma component and a sarcomatous component, and the background liver showed nodular hyperplasia. Immunohistochemical examination of the mass revealed positive staining for CD34, CK19, and pancytokeratin AE1/AE3, and negative staining for CA19, hepatocytes, AFP, HMBE-1, G3, TG, TTF1, and CK5/6.

FINAL DIAGNOSIS

Based on the histological and immunohistochemical findings, the tumor was diagnosed as an intrahepatic less differentiated sarcomatoid cholangiocarcinoma.

TREATMENT

The patient underwent radical hepatectomy. After surgery, he was treated with anti-infection agents, rehydration, and symptomatic treatment.

OUTCOME AND FOLLOW-UP

The overall duration of follow-up was only 2.5 mo as the patient passed away.

DISCUSSION

Sarcomatous changes that occur in cholangiocarcinoma are considered rare and the World Health Organization[3] classification of tumors defines sarcomatous intrahepatic cholangiocarcinoma (SICC) as a cholangiocarcinoma with spindle cell areas resembling spindle cell sarcoma or fibrosarcoma or with features of malignant fibrous histiocytoma. At present, the specific pathological mechanism of cholangiocarcinoma with sarcomatosis has not been clarified. Clinical manifestations as well as radiological imaging of this tumor are still limited.

An internet research was carried out using the search engines PubMed and Google with the keywords: "liver", "sarcomatous", "sarcomatoid", "sarcomatosis" and "cholangiocarcinoma". References of related literature were also reviewed to identify any other potentially relevant publications. After detailed search and analysis, 43 non-repeated cases of SICC were identified in 20 published studies[1,2,4-20].

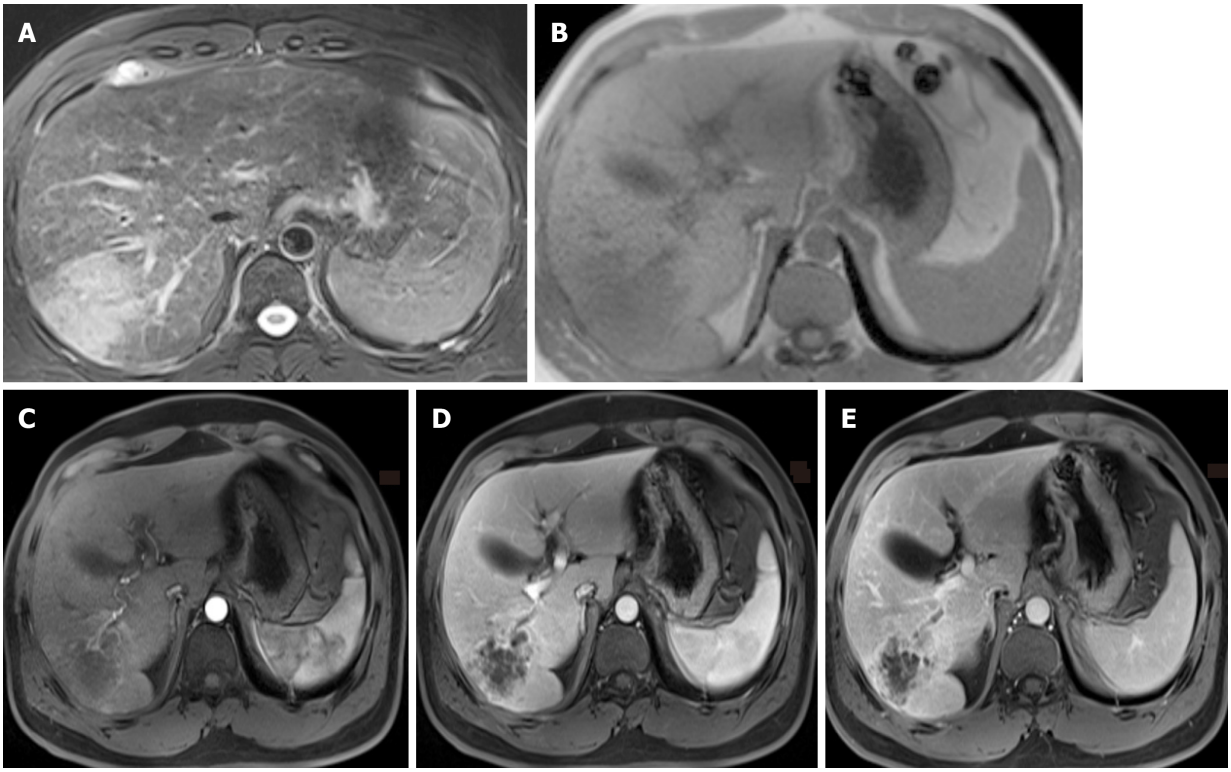

Figure 1 Magnetic resonance imaging results. A-B: There is an abnormal lesion occupying the right lobe of the liver, hyperintense on non-contrast T2 weighted images and hypointense on non-contrast T1 weighted images; C-E: A honeycomb-like structure and persistent enhancement with slight transient hepatic parenchymal enhancement around it and adjacent proximal bile duct dilatation with enhancement of the wall on contrast-enhanced T1 weighted images were observed.

The following tables (Tables 1-3) present a summary of the previous English-language literature with the addition of the present case. The clinical features of SICC were non-specific. Abdominal pain and fever were the most common complaints in patients. The age of the patients ranged from 37 to 87 years (median age, 61.5 years) with SICC being more commonly observed in men. This is consistent with the age and sex of the patient in the current study. Similar to typical cholangiocarcinoma, SICC usually occurs in the left lobe of the liver.

Radiologic imaging of SICC is still limited. It usually shows a clear low or mixed echo mass on ultrasound[21]. CT examination revealed low density lesions, clear or unclear, and sometimes with intra-tumor hemorrhage[15]. Most lesions had enhanced peripheral areas in the arterial phase and gradually filled in the central region. In our case, on CT analysis, the lesion was seen as a patchy low-density mass with an unclear boundary. Heterogeneous enhancement was observed in peripheral areas, with no obvious enhancement in the inner necrosis area. In addition, there was slight THPE around the mass. As a result, it is difficult to distinguish SICC from an atypical liver abscess. It was reported that satellite foci may appear in SICC; and there is a certain relationship between satellite nodules and SICC[14]. In the case reported herein, no daughter nodules were found. On magnetic resonance imaging, the lesion had low signal on T1WI and high signal on T2WI. After enhancement, a honeycomb-like structure and persistent enhancement with slight THPE and adjacent proximal bile duct dilatation with enhancement of the wall were observed. In addition, blood vessels were observed entering the lesion, which was also considered a distinguishing feature. The SICC involved multiple cystic changes accompanied by fibrous septum and was inhomogeneous and hyperintense in the center on DWI mapping, which was similar to atypical liver abscess on DWI mapping[18]. Unfortunately, our patient had no evidence of diffusion restriction. Abdominal angiography showed that the tumor was supplied by the hepatic artery and was considered anemic[7], which was consistent with the results after enhancement.

The degree of SICC malignancy is higher than that of traditional cholangiocarcinoma. The general prognosis for this malignancy is 3 mo[1]. It may be associated with an intrinsic sarcoma-like component, with high invasiveness. Most of these tumors are removed by surgery, and there is no unified comprehensive treatment for them. A previous study reported that surgery and postoperative treatment of a combination of gemcitabine and cisplatin adjuvant therapy can improve the prognosis of patients, and survival can be as long as 29 mo[13]. However,

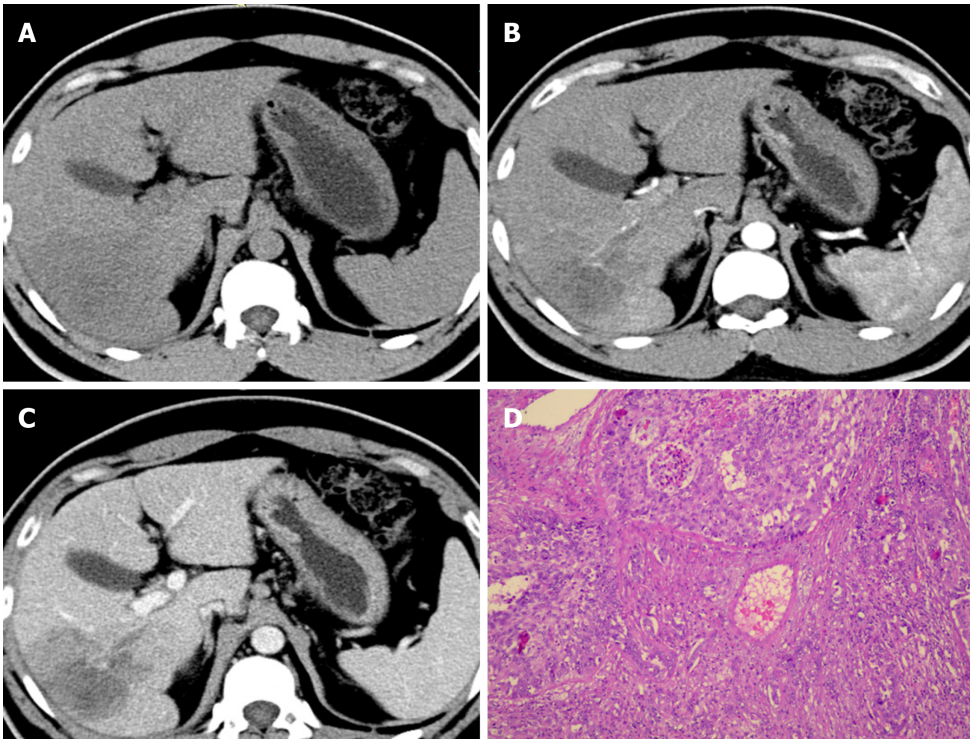

Figure 2 Computed tomography images and histopathologic findings (100 ×). A-C: The right lobe of the liver shows a patchy low-density shadow of approximately 7.0 cm × 5.6 cm with heterogeneous enhancement on magnetic resonance imaging; D: The cells are spindle to round in shape, with scanty cytoplasm and hyperchromatic nuclei, forming nests and microglandular structures.

the effectiveness of chemotherapy remains unclear. According to the survival curve (Figure 3), using the Kaplan-Meier method, in 44 patients with SICC reported in the literature and our case, the survival rate of patients who underwent surgery was significantly higher than those without surgery. The present patient did not undergo chemotherapy or radiotherapy after surgery, only supportive treatment. This was likely a contributing factor in his postoperative survival time of only 2.5 mo.

CONCLUSION

In summary, the clinical and radiological features of SICC are non-specific. The usual symptoms are abdominal pain and fever and the imaging features are hypovascularity and progressive enhancement. SICC can present as a multilocular cyst on radiological images and it is necessary to distinguish it from an atypical abscess. It is very difficult to diagnose SICC accurately prior to surgery. The last necessary preoperative biopsy is used for identification. Pathological diagnosis is the gold standard for diagnosing SICC. We report a case of SICC mimicking liver abscess and previous reports were reviewed to increase our understanding of the clinical and imaging evidence of SICC in order to improve the preoperative diagnostic ability for radiologists as well as surgeons.

Table 1 Summary of sarcomatous intrahepatic cholangiocarcinoma reported in English-language literature from 1992 to 2019

Ref.	Case No.	Age/sex	Clinical symptom	Location	Size (cm)	Treatment	Outcome (mo)
Nakajima *et al*[1]	1	84/F	Anorexia, jaundice, abdominal pain	Hepatic hilum	3.5	None	Died 3
	2	45/F	Fever	Right lobe	14.0	Surgery	Died 4.5
	3	73/F	Abdominal mass	Left lobe	7.0	Chemotherapy	Died 5
	4	37/M	Abdominal discomfort, epigastralgia	Left lobe	10.0	None	Died 2.5
	5	64/M	Abdominal discomfort, nausea	Left lobe	7.5	TAE	Died 1
	6	52/M	Right hypochondralgia	Right lobe	7.5	TAE	Died 2
	7	69/M	Fever	Left lobe	10.0	Surgery	Well 36
Inoue *et al*[2]	8	61/M	Abdominal pain, distention	Left lobe	25.0	Surgery	Died 1.1
Haratake *et al*[4]	9	59/M	Fever, icterus, abdominal mass	Hepatic hilar	NA	Supportive	Died 1
Imazu *et al*[5]	10	77/M	Liver tumor	Left lobe	7.0	Surgery	NA
Honda *et al*[6]	11	61/F	Back pain	Right and Left lobe	NA	Supportive	Died 3.8
Matsuo *et al*[7]	12	77/F	Abdominal pain	Left lobe	4.3	Surgery	Died 5
Itamoto *et al*[8]	13	70/M	Fatigue, fever	Right lobe	8.0	Surgery	Well 9
Kaibori *et al*[9]	14	69/F	Fever, abdominal pain	Left lobe	20.0	Surgery	Died 3
Gupta *et al*[10]	15	50/M	Jaundice	Hepatic hilum	2.0	NA	NA
Lim *et al*[11]	16	41/F	Palpable epigastric mass	Left lobe	17.0	Surgery	Alive 2
Sato *et al*[12]	17	87/M	Elevated ductal enzyme levels	Left lobe	4.0	Supportive	Died 3
Malhotra *et al*[13]	18	60/F	Abdominal pain, abdominal mass	Left lobe	20.0	Surgery and chemotherapy	Well 29
Bilgin *et al*[14]	19	48/M	Abdominal pain, fatigue	Left lobe	13.0	Surgery and chemotherapy	Alive 12
Jung *et al*[15]	20	59/M	Pain, dizziness, mild fever	Right lobe	18.0	Surgery and chemotherapy	Well 8
Watanabe *et al*[16]	21	62/M	Liver tumor, jaundice	Hepatic hilum	4.5	Surgery and chemotherapy	Died 11
Kim *et al*[17]	22	67/M	Abdominal pain	Left lobe	6.0	Surgery	Alive 6
Shi *et al*[18]	23	55/M	NA	Right lobe	NA	Surgery	Died 2
	24	47/M	NA	Right lobe	NA	Surgery	Died 6
Zhang *et al*[19]	25	63/M	Abdominal pain	Left lobe	NA	Surgery	Well 4
Kim *et al*[20]	26	45/M	Abdominal pain	NA	7.5	Chemotherapy	Died 1.6
	27	67/M	Abdominal pain	NA	2.5	Chemotherapy	Died 4.9
	28	55/M	Abdominal pain, fever	NA	6.5	Chemotherapy	Died 4.3
	29	66/M	Abdominal pain, fever	NA	10.0	Supportive	Died 0.7
	30	56/M	Abdominal pain, fatigue	NA	8.0	Chemotherapy	Died 2.4
	31	66/M	Abdominal pain	NA	7.5	Chemotherapy	Died 4.2
	32	68/M	BWL, fatigue	NA	6.0	Supportive	Died 0.6
	33	55/M	Abdominal pain, fever	NA	8.5	Chemotherapy	Died 1
	34	49/M	Abdominal pain, fever	NA	9.5	Chemotherapy	F/U
	35	65/M	Abdominal pain	NA	9.5	Supportive	Died 0.5
	36	61/M	Abdominal pain	NA	5.0	*Viscum album*	Alive 12.7

Tsou *et al*[21]	37	77/F	Abdominal pain, palpable mass, BWL	Left lobe	14.0	None	Died 2
	38	62/M	Abdominal pain, BWL	Right lobe	3.0	Unknown	F/U
	39	59/M	Abdominal pain, palpable mass, BWL	Left lobe	11.0	None	Died 1
	40	63/M	Dyspnea, BWL	Right lobe	14.0	None	Died 0.3
	41	64/M	Back pain	Left lobe	11.0	Surgery	Died 2
	42	50/F	Abdominal pain	Right lobe	4.5	Surgery	Died 2
	43	69/F	Abdominal pain, fever	Left lobe	2.5	Surgery	Alive 48
Our case	44	43/M	Abdominal discomfort	Right lobe	7.0	Surgery	Died 2.5

F: Female; M: Male; BWL: Body weight loss; TAE: Transarterial embolization; NA: Not available; F/U: Lost to follow-up.

Table 2 Clinical characteristics of sarcomatous intrahepatic cholangiocarcinoma reported in the English-language reports and our case

Clinical characteristics	Number
Age (yr)[1]	60.8 ± 11.3, 61.5
Gender (Male/Female)	33:11 (3:1)
Clinical symptoms	
Abdominal pain/discomfort	26/44 (59.1%)
Fever	11/44 (25.0%)
Abdominal mass	6/44 (13.6%)
Jaundice	4/44 (9.0%)
Digestive symptoms	2/44 (4.5%)
Fatigue	4/44 (9.0%)
Back pain	2/44 (4.5%)
Body weight loss	4/44 (9.0%)
Dizziness	1/44 (2.3%)
None	2/44 (4.5%)

[1]Values for age are presented as means ± SD (n = 44) and median.

Table 3 Summary of radiological features of 44 patients

		Radiological features	Number
Location		Left lobe	18/44 (40.9%)
		Right lobe	10/44 (22.7%)
		Hepatic hilum	4/44 (9.0%)
		Multiple	1/44 (2.3%)
		Unknown	11/44 (25.0%)
Tumor size (cm)[1]			9.0 ± 5.2
US		Hypoechoic	6/23 (26.1%)
		Hyperechoic	3/23 (13.0%)
		Mixed-echoic	10/23 (45.5%)
		Unseen	1/23 (4.3%)
		Unknown	3/23 (13.0%)
CT	Plain	Low density	19/19 (100%)
	Enhancement	Enhanced peripheral areas (progressive enhancement to the central region)	22/23 (95.7%) (12/23) (52.2%)
		Without enhancement	1/23 (4.3%)
MRI	T1 WI	Iso/Low intensity	6/8 (75.0%)
	T2 WI	High intensity border	1/8 (12.5%)
		High and low intensity	5/8 (62.5%)
	Unknown		2/8 (25.0%)
	Enhancement	Enhanced peripheral areas (progressive enhancement to the central region)	2/8 (25.0%) (2/8) (25.0%)
	Unknown		6/8 (75.0%)
Angiography		Hypovascularity	1/3 (33.3%)
		Unknown	2/3 (66.7%)

[1]Values for tumor size are presented as means ± SD (n = 44). CT: Computed tomography; MRI: Magnetic resonance imaging.s

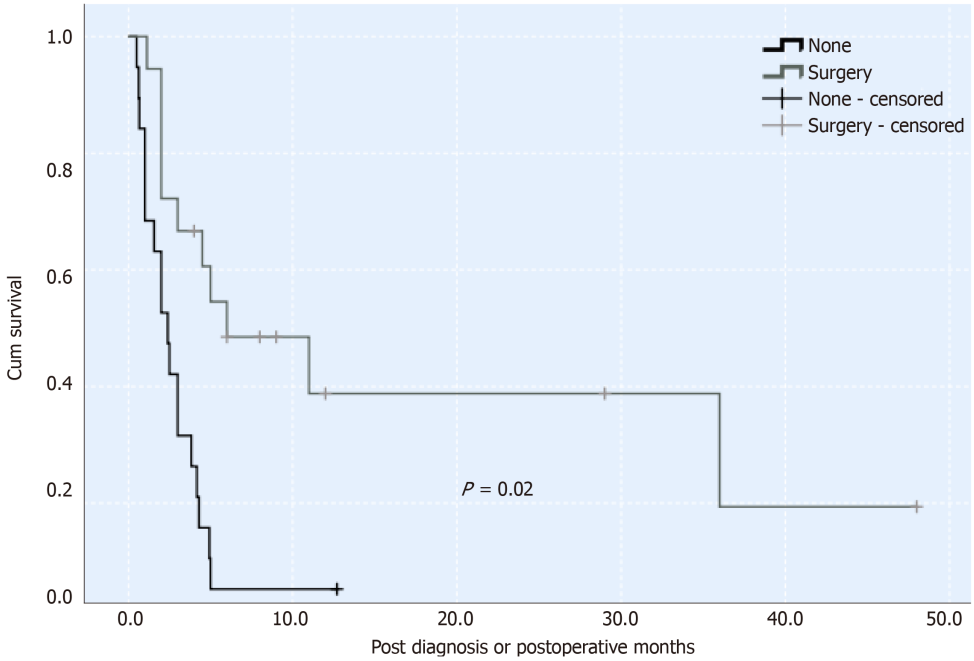

Figure 3 Survival rates of operated sarcomatous intrahepatic cholangiocarcinoma and non-operative sarcomatous intrahepatic cholangiocarcinoma. Survival rates of 44 cases of sarcomatous intrahepatic cholangiocarcinoma (SICC) calculated by Kaplan–Meier method [operated SICC (black solid line) and non-operative SICC (grey solid line)] are shown. The difference was estimated using the generalized log-rank method. *P* values of less than 0.05 are considered statistically significant.

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