Long-term follow-up of intrahepatic biliary cystadenoma and cystadenocarcinoma following hepatectomy

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Abstract

Backgroud: We present our experiences, particularly long term followup, of 20 patients who underwent operations for BCA or BCAC over the last 15 years in an effort to formulate more effective management strategies

Methods: Clinical data on patients with pathologically confirmed BCA or BCAC between June 2002 and May 2017 were retrospectively analyzed.

Results: Twenty patients were pathologically diagnosed with intrahepatic BCA (12) or BCAC (8); the mean ages for these 2 groups were 46.3 years and 62.0 years, respectively (P = 0.000). The main imaging feature for most BCA patients (83.3%) was cystic mass with septations. The main imaging features for BCAC were unilocular or unilocular cysts with papillary projections or solid portions. All BCAC patients received surgical treatment. No BCA patients developed recurrent cystadenoma after complete excision. Five (62.5 per cent) of the 8 BCAC patients died during a median follow-up time of 61.8 months. Three deaths were due to metastasis. The median follow-up of BCAC patients without a recurrence was 90.7months (range 31–152).

Conclusions: Factors associated with a higher likelihood of BCA or BCAC include being a middle-aged or older woman, having elevated serum CA19-9 levels, showing high-risk features on imaging, and having recurrent liver cyst(s). Radical excision is recommended to obtain long-term tumor-free survival.

Background

With current developments in cross-sectional imaging, increasing numbers of hepatic cystic lesions are being discovered. Intrahepatic Biliary cystadenomas (BCA) or biliary cystadenocarcinomas (BCAC) account for 5% of hepatic cysts [1]. Keen first reported BCA in 1892; Subsequently, Edmondson (1958) defined the pathognomonic pathologic features of BCA as a multilocular lesion lined by columnar epithelium with an accompanying densely cellular (“ovarian-like”) stroma[2]; and until now, fewer than BCA or BCAC cases worldwide have been reported in the literature[3]

Because of their rarity and a lack of specific diagnostic methods, diagnosis and treatment is often delayed and inappropriate. BCA are believed to be premalignant and may carry a risk of malignant transformation and recurrence; thus, accurately diagnosing and treating BCA is critical. We present our experiences, particularly long term followup, of 20 patients who underwent operations for BCA or BCAC over the last 15 years in an effort to formulate more effective management strategies.

Methods

Clinical data on patients with pathologically confirmed intrahepatic BCA or BCAC between June 2002 and May 2017 were analyzed retrospectively. Clinical data included demographic characteristics, symptoms, previous treatments, preoperative examinations, treatments and patient outcomes. Two pathologists

reviewed the pathological specimens from the operative resections. Follow-up data were obtained by reviewing the hospital records or by conducting telephone interviews. This study was approved by the Ethics Committee of Anhui Medical University (PJ2019-01-11).

Results

Twenty patients diagnosed with BCA or BCAC between June 2002 and May 2017 were identified by physicians at our institution and included in the study cohort. Their clinicopathological characteristics are summarized in Table 1. The patients were divided into 2 groups by pathological type: the BCA group (n = 12) and the BCAC group (n = 8). Patients with BCAC were older than those with BCA (62.0 years vs 46.3 years, $P < 0.001$). The median maximum diameter of all lesions was 107 mm (range 50 mm–200 mm). The BCA or BCAC imaging features are shown in Table 2.

Table 1
Patient characteristics

	Cystadenoma group (n = 12)	Cystadenocarcinoma group (n = 8)
Age (years)	46.3 (1–66)	62.0 (51–79)
Gender (M/F)	2/10	3/5
Past surgical history of BCTs	6 (50%)	0 (0)
Presenting symptoms		
Abdominal discomfort	8	4
Abdominal mass	1	2
Fever	0	1
Asymptomatic	3	2
Symptom duration (months)	18.7 (1–84)	12.0 (1–48)
HBsAg (+) (%)	3 (25.0%)	2 (25.0%)
CA19-9 ≥ 200 (U/ml) (%)	2 (22.2)	3 (37.5)
Operating time (min)	138.3 (115–195)	163.1 (125–215)
Location		
Left	7	7
Non-left	5	1
Tumor size (cm)	10.8 (6–18)	10.6 (5–20)
Pathology/Ovarian-like stroma	10 (83.3%)	6 (75.0%)
Adjuvant chemotherapy	0 (0)	3 (37.5%)

Table 2
High-risk features of intrahepatic biliary cystic tumors on imaging

	Cystadenoma group (n = 12)	Cystadenocarcinoma group (n = 8)
Preoperative imaging, n (%)		
Ultrasound	12 (100)	8 (100)
Contrast-enhanced CT	9 (75)	7 (87.5)
MRI	2 (16.7)	4 (50)
PET/CT	1(8.3)	1 (12.5)
Preoperative image finding feature, n (%)		
Septa	10 (83.3)	7 (87.5)
Mural nodularity or solid portion	2 (16.7)	7 (100)
Calcification	3 (25)	0 (0)
Hypervascular	1 (8.3)	7 (87.5)
Enhancement after contrast	3 (33.3)	7 (100)

CT: computed tomography; MRI: magnetic resonance imaging; PET/CT: positron emission tomography-computed tomography.

BCA subgroup

Nine patients visited the hospital for abdominal pain and discomfort or an abdominal mass. The youngest patient was 11 months old and was seen at the hospital for “an abdominal mass for 3 months” (Fig. 1A/B). The longest duration of an abdominal mass prior to final treatment was 7 years. Nine patients underwent CA19-9 testing, and of these, 2 had significantly high CA19-9 (881.9 U/mL and 261.6 U/mL). The typical imaging feature for most BCA patients (83.3%) was a cystic mass with septations (Fig. 1; Table 2). One underwent positron emission tomographic-computed tomographic imaging (PET/CT) because of her high CA19-9 and was found to have a benign cystic liver tumor.

Six patients (50%) had undergone 8 prior interventions, including percutaneous aspiration (1 patient) and laparoscopic or open unroofing (5 patients). All 12 patients underwent complete removal of the cystadenoma, including 6 cyst enucleations, 1 right hepatectomy, 2 left hepatectomies, and 3 hepatic left lateral lobectomies. Intraoperative pathological examination of frozen specimens was performed for 5 patients; 3 patients were diagnosed with BCA, and 2 were diagnosed with simple liver cysts. In 2 patients,

the cyst fluid was brown and cloudy, while in the remaining 10 patients, the cyst fluid was yellowish, mucinous, and transparent.

During follow-up, 2 patients had bilomas that resolved after percutaneous transhepatic drainage. Three BCA patients who previously underwent palliative treatment before March 2007 at our hospital (open drainage, n = 2; percutaneous transhepatic drainage, n = 1) relapsed after surgery and received a second surgery (complete tumor resection). During follow-up, no relapse occurred in the 12 patients who underwent complete cystadenoma removal.

BCAC subgroup

The BCAC patients' clinical data are summarized in Table 3. Patient #5 underwent cholecystectomy + choledocholithotomy + T-tube drainage 1 year before BCAC surgery, while the remaining 7 patients had no previous surgical history. The main imaging features seen with BCAC were locules or unilocular cysts with papillary projections or solid portions. The solid portions (walls, septations, papillary projections or solid portions) were enhanced after administering an intravenous contrast (Figs. 2 and 3; Table 2). For patient #3, PET/CT showed a left hepatic lobe lesion with abnormally high phosphogluconate dehydrogenase (PDG) metabolism, suggesting biliary carcinoma (Fig. 2C). The longest duration of an abdominal mass prior to final treatment was 4 years.

Table 3
Clinical features and prognostic data of cystadenocarcinoma

Case	Age	Sex	CA19-9 (U/ml)	Size (cm)	Operation	Margin	Follow-up
1	60	Female	13.2	12	1	R0	FOD, 76 mo. No recurrence
2	51	Female	24.1	7	2	R1	DOD, 33 mo. Intrahepatic recurrence in 3 mo., treated with TACE
3	60	Female	1000.0	14	1	R0	DOD, 57 mo. Intrahepatic recurrence and regional lymph node metastasis in the 48 mo., treated with PTCD
4	63	Male	10.30	9	1	R0	FOD, 104 mo. No recurrence
5	58	Female	0.6	5	3	R1	DOD, 36 mo. Intrahepatic recurrence and lymph node metastasis 32 mo.
6	65	Male	1000.0	10	4	R0	Died from biliary infection and biliary bleeding in 5 mo.
7	60	Male	3.45	8	1	R0	DOD, 152 mo. No recurrence
8	70	Female	209.3	20	1	R0	FOD, 31 mo. No recurrence

Operation Legend: 1, left hepatectomy; 2, cyst enucleation + choledocholithotomy + T-tube drainage; 3, cyst enucleation; 4, right posterior lobectomy + choledocholithotomy + T-tube drainage; DOD, died of disease; FOD, free of disease; TACE, transcatheter arterial chemoembolization; PTCD,percutaneous transhepatic cholangial drainag.

All 8 patients underwent surgery (Table 3). Patient #5 had a tumor thrombus in the left hepatic duct. Patients #2 and #6 had intrahepatic biliary stones, and patient #2 had yellowish ascites (300 mL). Patients #2 and #5 experienced iatrogenic tumor rupture with a positive biliary surgical margin (R1). No intraoperative pathological examinations (frozen specimens) were performed.

Detailed follow-up information is shown Table 3. Follow-up times ranged from 5 to 152 months, with a median follow-up time of 61.8 months (Table 3). During follow-up, patient #1 (Fig. 3) had a biloma 2 months after a left hepatectomy, which resolved after percutaneous transhepatic drainage. Patients #2, #5 and #7 underwent fluorouracil-based chemotherapy after surgery.

Five (62.5 per cent) of the 8 BCAC patients died during follow-up. Three deaths (patients #2, #3, and #5) were due to metastasis 33 months, 57 months and 36 months after surgery. Two patients (Patient #6 and #7)died from biliary bleeding and multiple organ failure without tumor recurrence at 5 and 152 months

respectively after surgery.The remaining 3 patients (with radical resections) remained alive and recurrence-free at the end of the follow-up period.

Discussion

BCA or BCAC are a subset of cystic lesions that comprise < 5% of all hepatic cysts; BCA incidence is higher in women, but no significant gender difference is observed in BCAC incidence. On average, BCAC patients are 55 years old, approximately 10 years older than BCA patients [1]. In this study, the male to female BCA patient ratio was 1:5; for BCAC, the ratio was 3:5. The average age of onset was 46.2 years in BCA patients and 62.0 years in BCAC patients, with a significant between-group age difference, consistent with other literature reports. The cause of BCA or BCAC is unknown. In this study, an 11-month-old child had a giant BCA, confirming some researchers' opinions that BCA is a congenital condition associated with congenital biliary malformation [4]. BCA or BCAC grow slowly. Thomas et al. [5] showed that BCA usually persisted an average of 3 years before the initial hospital visit; 1 BCAC patient (with a suspected benign hepatic tumor before surgery) was followed for 10 years before surgery [6]. In this study, BCA or BCAC persisted an average of 16.7 ± 23.7 months (up to 8 years) before the initial visit. BCA or BCAC lack specific clinical symptoms and may cause bloating, abdominal pain, and jaundice because of tumor oppression of the abdominal organs. In this study, patient #6 had high fever before surgery because of intrahepatic biliary stones and ensuing biliary infection.

For BCA or BCAC, laboratory tests are nonspecific. Alpha-fetoprotein (AFP) is usually normal, and the serum and cyst fluid CA19-9 levels help differentiate cystic neoplasm and simple liver cysts, but not BCA or BCAC [7–8]. This study showed that the serum AFP was normal in all 20 patients, but the CA19-9 was significantly increased in 2 BCA patients and 3 BCAC patients. Before surgery, BCA or BCAC are diagnosed mainly via imaging. Typical imaging findings include mono- or multilobe cystic lesions and cystic or solid masses, with partitions or papillary projections. Enhanced scans show persistent enhancement in the posterior cystic wall, partitions, and papillary projections. However, these findings are observed in both BCA and BCAC [3, 9]. Arnaoutakis et al. [3] retrospectively analyzed the imaging findings of 248 BCT cases from multiple centers worldwide and found that 81.5% of the BCAC cases and 79.2% of the BCA cases (no significant between-group difference) had 1 of the following at-risk imaging signs: multilobes, partitions, papillary projections, calcification, high-flow signals, enhancement, or biliary dilatation. Moreover, Arnaoutakis et al. [2] noted that for BCACs with 1 of the imaging findings described above, the rate of negative prediction was high (91%), while the rate of positive prediction was only 11%. Among the 8 cases of BCAC, 5 presented a cystic mass with partitions and papillary projects or a high flow signal; and 3 presented a solid mass with a high flow signal. However, the diagnosis rate from preoperative imaging was low, which may be related to some radiologists' lack of understanding of these conditions.

BCA are often misdiagnosed as other hepatic cystic lesions, especially simple liver cysts, increasing the rate of repeat surgery. Thomas et al [4] retrospectively analyzed 18 BCA cases treated at the Vanderbilt

University School of Medicine and showed that before surgery, all patients had been seen by 1 to 4 surgeons and several physicians, of which, 8 patients underwent a total of 20 palliative surgeries. This study showed that among BCA patients, 50% had previously undergone palliative treatment before complete tumor resection. Thus, BCA or BCAC must be differentiated from complex and atypical liver cysts to improve treatment and determine patient outcomes. In our experience, the following patients should be carefully monitored: (1) middle-aged and elderly female patients with liver cysts; (2) patients whose imaging findings include mono- or multilobe cystic tumors with papillary projections or partitions; (3) patients with postoperative relapses and “liver cysts” with high CA19-9 levels; and (4) patients who present with cystic jelly-like viscous fluid, papillary projections, or partitions during their operations.

Grossly, BCA are generally lobular, multilocular cystic lesions with partitions or papillary projections. Histologically, the World Health Organization (WHO) defines ovarian-type stroma as a necessary condition for diagnosing hepatobiliary mucinous cystic neoplasms[2, 4], but this is not widely applied today[3, 8, 10]. All patients enrolled in studies showing a 100% ovarian-type stroma-positive rate were women[11]. Vogt et al[12] reported 18 BCA patients, all of whom were women, and only 55.5% were positive for ovarian-type stroma. One of the most comprehensive studies[3], including 248 cases, noted that only one-third of BCA/BCAC lesions presented evidence of ovarian-like stroma. Two large studies on BCAC included 27 and 18 patients with an approximately 1:1 male:female ratio, and only 30% of patients were positive for ovarian-type stroma[3, 10]. Therefore, some researchers conclude that ovarian-type stroma (+) may be a typical feature and a prognostic predictor of BCA. Devaney[10] reported 30 cases of mesenchymal-associated BCAC; the patients were followed for 4 to 8 years and none died. Conversely, the mean survival was 3 years for BCAC patients without mesenchymal stroma. In this study, patient #7, a man with mesenchymal-associated BCAC, achieved tumor-free survival for 139 months after radical resection.

Differentiating benign and malignant cystic neoplasm before surgery is difficult, as is differentiating cystic neoplasm and simple hepatic cysts or benign and malignant cystic neoplasm using intraoperative frozen pathology[3, 13]. Martel[13] showed that intraoperative frozen pathology differentiated cystic neoplasm from simple hepatic cysts in only 33.3% of cases. Vogt [12] reported a case of highly suspected BCA or BCAC, where fast intraoperative frozen pathology suggested simple hepatic cysts for six specimens. This suggests that in patients for whom BCA or BCAC is highly suspected before surgery, surgeons must consider factors other than intraoperative frozen pathology when deciding the surgical approach. BCA can become malignant, with an extremely high local relapse rate in cases of noncomplete or partial resection; thus, radical resection is the preferred treatment. Palliative surgical approaches, such as lesion puncture (to withdraw fluid), injection of hardening agents, internal drainage, open drainage, and partial resection, are contraindicated and should be avoided [3, 5, 81, 14]. The BCA relapse rate is 48.6% after open drainage and partial resection but only 10% after an extensive hepatectomy [3]. In this study, all 6 BCA patients relapsed after palliative treatment. After relapse, patient outcomes remained good after a second surgery. Arnaoutakis et al. [3] retrospectively analyzed 33 BCA or BCAC patients who relapsed and underwent a second surgery. These authors showed that 11 underwent partial hepatectomies, and 22

underwent semi-hepatectomies or expanded semi-hepatectomies. The patients were followed for an average of 18.5 months, and 78.8% achieved tumor-free survival.

BCAC is a relatively inert malignant tumor with a good prognosis after complete resection. Arnaoutakis et al. [3] retrospectively analyzed 248 BCA or BCAC cases and showed that the overall survival rates were 95.0%, 86.8%, and 84.2% at 1, 3, and 5 years after BCT resection, respectively. For BCAC patients in the study, the median survival time was 8.4 years[3]. The two patients with iatrogenic tumor rupture and positive biliary surgical margins died of carcinomatosis, while the remaining 4 patients achieved long-term survival after radical resection. These results showed that tumor rupture is an important negative prognostic factor of recurrence-free survival and overall survival, and complete resection (R0 resection) is the key for long-term survival.

Conclusion

Middle-aged or older women, patients with elevated serum CA19-9 levels, patients with high-risk features on imaging, and patients with recurrent liver cyst(s) have higher possibilities of biliary cystic tumors. Radical resection is the preferred treatment, which helps to ensure a good overall prognosis for BCA or BCAC patients. Nevertheless, some patients may relapse and must be followed up as scheduled. In relapse cases, patients may still undergo a second or repeat surgery.

Additional File

Video S1. Left hemihepatectomy for Primary intrahepatic billary cystadenocarcinoma (Patient #1) (MP4; 34994 KB)

Abbreviations

BCA: biliary cystadenoma; BCAC: cystadenocarcinoma; CT: computed tomography; MRI: magnetic resonance imaging; PET/CT: positron emission tomography-computed tomography; DOD, died of disease; FOD, free of disease; TACE, transcatheter arterial chemoembolization; PTCD,percutaneous transhepatic cholangial drainag

Declarations

Ethics approval and consent to participate

The project has been approved by the Committee on Medical Ethics of The First Affiliated Hospital of Anhui Medical University. The reference number is “PJ2019-01-11”. written

consent was requested and obtained from all patients at the time of admission.

Consent for publication

Informed consent was obtained from the patients for the publication of individuals' clinical details, any accompanying images and supplementary video at the time of admission.

Availability of data and materials

The datasets used and analysed during the current study are available from the corresponding author on reasonable request.

Conflict-of-interest statement

All authors declare no conflicts-of-interest related to this article.

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Author contributions

All authors helped to perform the research; CJM manuscript writing, performing data analysis, drafting conception and design; GW manuscript writing and drafting conception and design; ZYJ, XK, WGB and GXP acquired the data, provided technical support and performed procedures. LFB contributed to drafting conception and design, performing procedures, writing the manuscript and data collection.

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Not applicable

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Figures

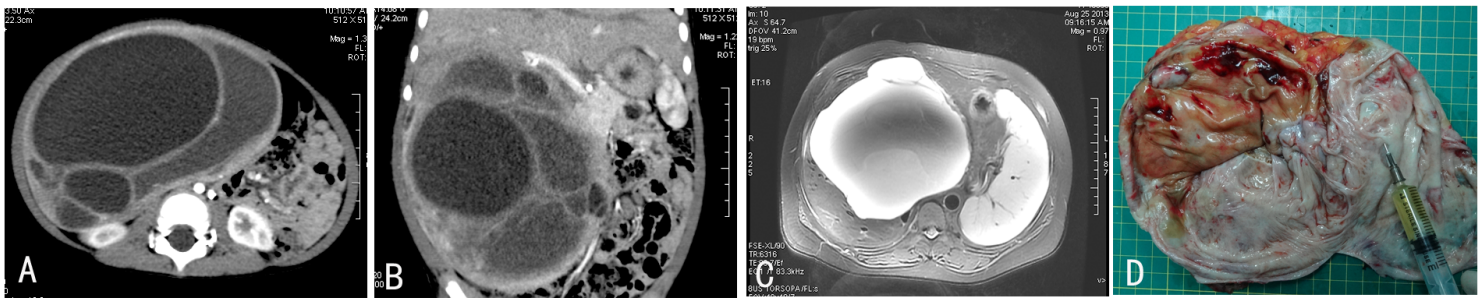

Figure 1

Biliary cystadenoma. (A/B) Biliary cystadenoma in an 11-month-old male patient. (A) Axial and (B) coronal CT images in the portal venous phase show multilocular cystic lesions with internal septations. (C/D) Biliary cystadenoma of a 44-year-old female patient with a history of liver cysts who had undergone 2 fenestration operations. (C) Coronal T2-weighted magnetic resonance image (MRI) shows a solitary hyperintense lesion with internal septations. (D) The internal view with the open cyst cavity shows a solitary cyst with internal septations and yellowish cystic fluid.

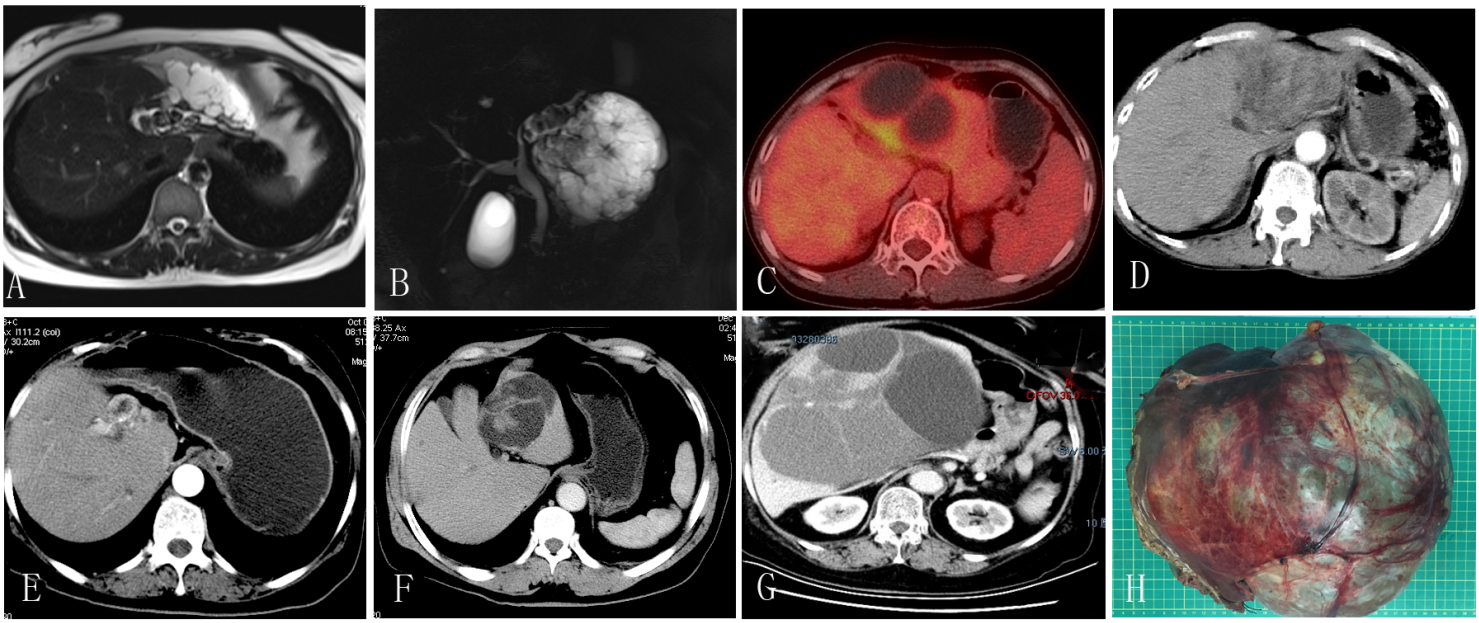

Figure 2

Biliary cystadenocarcinoma. (A) T2-weighted MRI image and (B) magnetic resonance cholangiopancreatography (MRCP) show a multilocular-solid cyst with irregular thick walls and hepatolithiasis in patient #2. (C) PET/CT of patient #3 shows a left hepatic lobe multilocular-solid cyst with abnormally high PDG metabolism. (D) Computed tomography of the liver in the portal venous phase shows a unilocular-solid cyst in the left lobe in patient #4. (E) After operations over 2 years, computed tomography of the liver in the portal venous phase shows a multiloculated cystic mass with associated ductal dilation in patient #5. (F) Axial CT images in the portal venous phase shows an exogenous, solitary cystic mass with convex papillate in patient #7. (G) Computed tomography of the liver shows a multilocular cyst in the left lobe with internal septations and papillary projections in patient #8. (H) External view of the resected cystadenocarcinoma in patient #8.

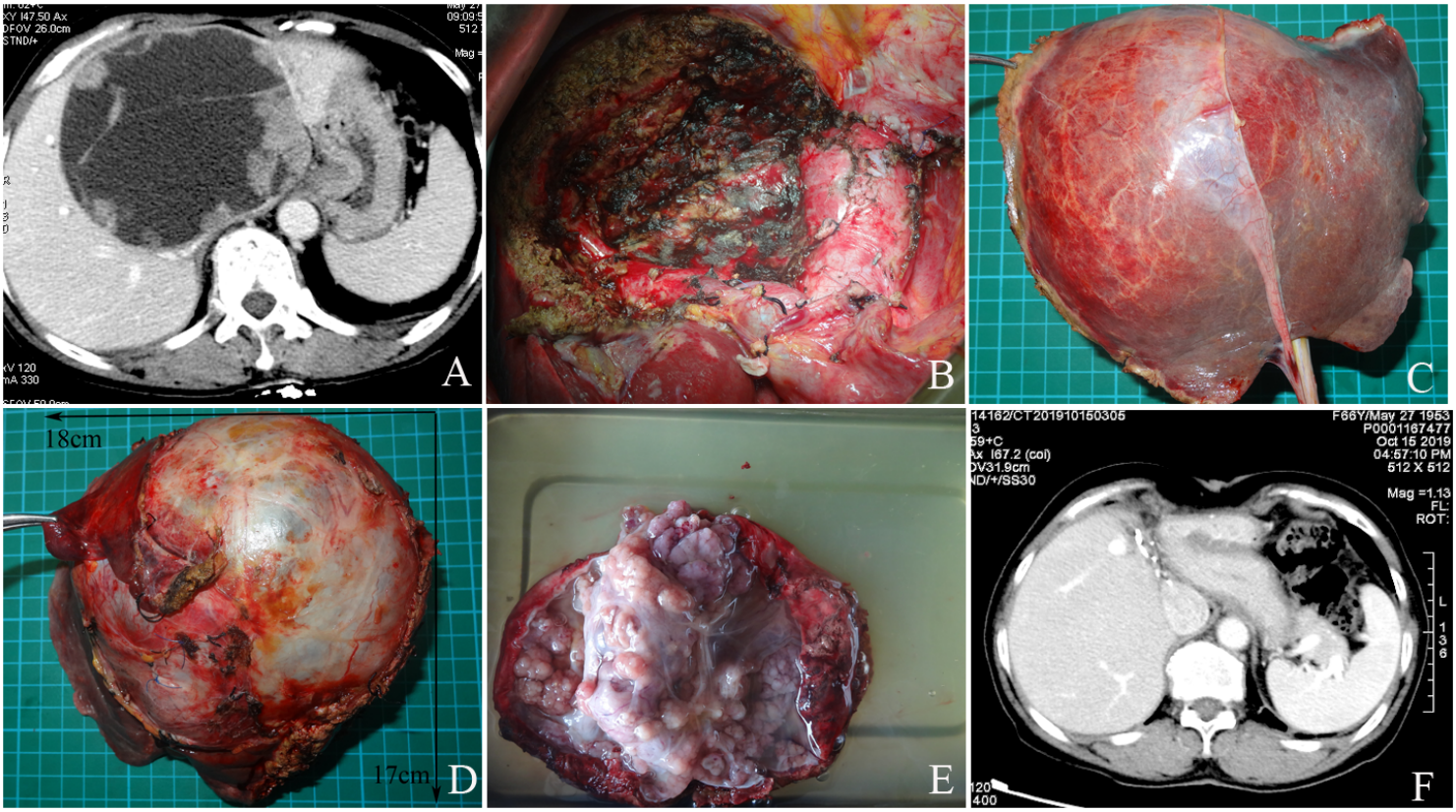

Figure 3

Biliary cystadenocarcinoma in patient #1. (A) Computed tomography of the liver in the portal venous phase shows a solitary cyst in the left lobe with internal septations and enhanced mural nodules. (B) Remaining right lobe of the liver after extended left hepatectomy. (C) External view of the resected specimen, including the resected cystadenocarcinoma, left, and the Spiegel lobe of the liver. (D) Internal view with the open cyst cavity shows a solitary cyst with a thickened wall, internal septations, papillary mural nodules and yellowish cystic fluid. (E) Histopathology showing cystadenocarcinoma with complex papillary architecture and extensive cytologic atypia (hematoxylin and eosin stain, original magnification ×400). (F) CT image of the liver after resection (63 months) shows no tumor recurrence.

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- Patient1.mp4