Case Report

Sarcomatoid intrahepatic cholangiocarcinoma in a patient with poor prognosis: a case report and literature review

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Abstract
Sarcomatoid intrahepatic cholangiocarcinoma (S-iCCA) is a rare histological variant of intrahepatic cholangiocarcinoma (iCCA). The diagnosis of S-iCCA is based on histopathological and immunohistochemical examinations, and S-iCCA often has a poorer prognosis than that of ordinary iCCA. In this article, we present the case of a 64-year-old man with S-iCCA who presented with intermittent right upper abdominal pain. The aim of this case report and literature review is to strengthen the understanding of S-iCCA among clinicians and reduce the incidence of missed clinical diagnoses.

Keywords
Sarcomatoid change, sarcomatoid intrahepatic cholangiocarcinoma, poor prognosis, case report, hepatobiliary malignancy, abdominal pain

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Introduction
Epithelial tumors with sarcomatoid changes are rare neoplasms that have been reported in various sites, including the upper digestive tract, lung, pancreas, skin, breast, thyroid, uterus, urinary tract, and gallbladder.1,2 Sarcomatoid intrahepatic cholangiocarcinoma (S-iCCA) is defined by the World Health Organization as

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intrahepatic cholangiocarcinoma (iCCA) with sarcomatoid changes. The mechanism of its pathogenesis is still unknown. It has been reported that sarcomatoid hepatocellular carcinoma (HCC) may be associated with preoperative anticancer treatments, such as transcatheter arterial chemoembolization, radiofrequency ablation, or percutaneous ethanol injection. However, there is no relevant study demonstrating the association between anticancer treatments and S-iCCA. Confirmation by biopsy and immunohistochemical staining is critical to establish a definitive diagnosis of S-iCCA. Histological and immunohistochemical examinations of S-iCCA typically show a malignant neoplasm with both carcinomatous and sarcomatous components and positive expression of epithelial and mesenchymal molecular markers. At present, the main treatment option is surgical resection; however, patients still have a relatively poor prognosis. There are a few reports on S-iCCA worldwide, and most of them are individual cases. This case report and review of relevant literature provide useful information to improve the awareness of S-iCCA among clinicians, reduce missed diagnoses, and achieve an accurate diagnosis and treatment.

**Case report**

The patient was a 64-year-old Chinese man admitted to our hospital for intermittent right upper abdominal pain. Physical examination showed tenderness and rebound pain in the right upper abdomen. Serological examinations revealed a slightly elevated gamma-glutamyl transpeptidase level at 119.3 U/L (normal range: 10–60 U/L), an elevated serum carbohydrate antigen 19–9 (CA19-9) level at 351.74 U/mL (normal range: 0–37.0 U/mL), and an elevated 24–2 level at 86.55 U/mL (normal range: 0–20.0 U/mL). His serum carcinoembryonic antigen and alpha-fetoprotein levels were within normal limits. A computed tomography scan of the entire abdomen plus three-stage enhancement (Figure 1) showed multiple calcifications in the left inner lobe of the liver and multiple calculi of intrahepatic and extrahepatic bile ducts, followed by dilatation of intrahepatic and extrahepatic bile ducts (part of the left outer lobe of the liver had expanded to the thickness of the bile ducts, and the contrast enhancement was obvious).

The preoperative diagnoses were space-occupying lesions in the left lateral lobe of the liver and extrahepatic and intrahepatic cholangiolithiasis. No related lymph node or distant metastasis was found. Hepatic left lateral lobectomy, cholecystectomy, exploration and lithotomy of the biliary tract, and T tube drainage were performed. Histological examination (Figure 2) of the liver tissue demonstrated the presence of an S-iCCA (2.0 × 1.8 × 1.7 cm) confined to the hepatic capsule; no evidence of cancer infiltration was found at the margin of hepatectomy, in vessels, or in nerves. A separate inspection (embolus) revealed the presence of cancer tissue. The tumor stage was determined to be T2aN0M0 based on the 7th edition of the American Joint Committee on Cancer TNM staging system. Immunohistochemical examination of the neoplasm showed positive staining for cytokeratin (CK)8, CK-pan, and vimentin and negative staining for CK7, CK20, and hepatocyte paraffin 1 (HepPar-1). The Ki-67 proliferation index was approximately 60%. Based on these histopathological and immunohistochemical findings, a definitive diagnosis of S-iCCA was determined. The overall follow-up duration was 3 months, and the patient died 3 months after surgery.

**Discussion**

S-iCCA is a rare histological subtype of iCCA and has only been reported in the
English literature in case reports or series. Its prevalence is unknown, but it is reported to account for 4.5% of cholangiocarcinoma cases\(^8\) and less than 1% of hepatobiliary system malignancies.\(^9\) As for the pathogenesis, researchers have proposed that anti-cancer treatments might lead to the development of sarcomatoid changes or

![Figure 1. Axial computed tomography findings in the described patient with sarcomatoid intrahepatic cholangiocarcinoma. (a) and (b) Arterial phase reveals part of the left outer lobe of the liver that has expanded to the thickness of the bile ducts, and strengthening is obvious. (c) and (d) The lesion shows no contrast enhancement in venous or late phase reveals.](image1)

![Figure 2. Histologic findings of liver biopsy in the described patient with sarcomatoid intrahepatic cholangiocarcinoma. (a) and (b) Hematoxylin and eosin staining, 10\(\times\) and 40\(\times\), respectively. Immunohistochemistry (20\(\times\)) for CK-pan (c), HepPar1 (d), and vimentin (e). CK-pan, pan-cytokeratin; HepPar1: hepatocyte paraffin 1.](image2)
accelerate the transformation of epithelial-derived cells into sarcoma cells.\(^{10}\) However, there are no reports regarding the relationship between S-iCCA and anticancer therapy.\(^{8}\)

Abdominal pain is the most common clinical symptom of S-iCCA. According to previous studies,\(^{9,11,12}\) serum CA19-9 and carcinoembryonic antigen (CEA) may not be sensitive enough for the diagnosis of S-iCCA. Of note, a low echogenic liver mass on ultrasound showing hypoattenuation and peripheral region enhancement after contrast injection on a computed tomography scan\(^{9}\) are common characteristics of S-iCCA, similar to ordinary iCCA.\(^{7}\) Therefore, it may be difficult to distinguish S-iCCA from ordinary iCCA using radiological imaging. A definitive diagnosis of S-iCCA can be determined by biopsy.

To review the known characteristics of S-iCCA, we searched PubMed using the keywords “liver”, “sarcomatous”, “sarcomatoid”, and “cholangiocarcinoma”. The clinical features of 46 cases are summarized in Table 1 (including the present case). Among these patients, 32 (69.6%) were male, 14 (30.4%) were female, and the mean age was 62 (range: 37–87) years. The mean tumor size was 8.0 (range: 2.0–22.0) cm. The tumor location was recorded in 21 patients, most frequently in the left lobe of the liver (13 patients; 61.9%), followed by the right lobe (7 patients; 33.3%) and the hepatic hilum (1 patient; 4.8%). Overall, 36 (78.3%) patients had one tumor, and 10 (21.7%) patients had multiple tumors. The initial radiologic impressions were recorded in 25 patients, with 18 (72.0%) characterized as HCC, 1 (4.0%) as lymphoma, 3 (12.0%) as hepatic abscesses, and 3 (12.0%) as hepatic space-occupying lesions. The laboratory findings are shown in Table 2. CA19-9 levels were elevated in 15 patients and normal in 20 patients; CEA was elevated in 3 patients and normal in 19 patients. Detailed immunohistochemistry results were available for 33 patients, which are shown in Table 3. Overall, tumors were positive for CK in 30 (90.9%) patients and positive for vimentin in 27 (81.8%) patients, which was considerably helpful to arrive at the final diagnosis of S-iCCA. Among the 46 S-iCCA patients who had treatment information available (Table 4), 22 (47.8%) underwent surgical treatment, 13 (28.3%) underwent chemotherapy or radiotherapy, 2 (4.3%) underwent transcatheter arterial embolization, and 9 (19.6%) remained untreated.

The diagnosis of the tumor is established by histopathological and immunohistochemical examinations. S-iCCA does have some unique histopathological and molecular cytogenetic patterns. Histopathological analyses of S-iCCA show the coexistence of adenocarcinoma cells with differentiated and sarcomatoid cells, which are spindle-shaped and arranged in bundles or weaves. Immunohistochemistry reveals that S-iCCA tumors are positive for both epithelial cholangiogenic tumor markers (CK7, CK8) and the mesenchymal tumor marker vimentin and negative for HepPar-1.\(^{4,8,11,12}\) As a marker of hepatocytes, HepPar-1 provides useful diagnostic information in distinguishing HCC from cholangiocarcinoma and metastatic carcinoma in the liver. In addition, our patient (described above) was positive for CK-pan, CK-8, and vimentin and negative for CK20 and HepPar-1, which are consistent with our diagnosis.

At present, there are no relevant guidelines for determining the prognosis and survival of patients with S-iCCA, and radical liver resection is the only available treatment. In published case reports, the median survival of patients with S-iCCA who underwent surgical resection was 11 months, which is comparable with that in patients with ordinary iCCA who did not undergo surgery (8 months).\(^{4}\) The median survival of patients with S-iCCA who did
Table 1. Clinical characteristics of S-iCCA reported in the English-language literature.

| Case no. | Age (y) | Sex | Location      | Tumor size (cm) | Number of tumors | Initial radiologic impression |
|----------|---------|-----|---------------|-----------------|------------------|-----------------------------|
| Kim et al\(^9\) | 1 45 | M | N/A | 7.5 | Multiple | HCC |
|         | 2 67 | M | N/A | 2.5 | Single | HCC |
|         | 3 55 | M | N/A | 6.5 | Multiple | IHCC |
|         | 4 66 | M | N/A | 10 | Single | Hepatic abscess |
|         | 5 56 | M | N/A | 8 | Single | HCC |
|         | 6 66 | F | N/A | 7.5 | Single | IHCC |
|         | 7 68 | M | N/A | 6 | Single | HCC |
|         | 8 55 | F | N/A | 8.5 | Multiple | IHCC |
|         | 9 49 | M | N/A | 9.5 | Multiple | Lymphoma |
|         | 10 65 | M | N/A | 9.5 | Multiple | IHCC |
|         | 11 61 | M | N/A | 5 | Single | IHCC |
| Sintra et al\(^18\) | 12 N/A | M | Right | 10 | Single | Hepatic carcinoma |
| Sasaki et al\(^19\) | 13 79 | M | Left | 8 | Multiple | Hepatic mass |
| Haratake et al\(^20\) | 14 59 | M | Right | Fist-sized | Multiple | Hepatic abscess |
| Nakajima et al\(^21\) | 15 84 | F | Hepatic hilum | 3.5 | Single | N/A |
|         | 16 43 | F | Right | 14 | Single | N/A |
|         | 17 73 | F | Left | 7 | Single | N/A |
|         | 18 37 | M | Left | 10 | Single | N/A |
|         | 19 64 | M | Left | 7.5 | Single | N/A |
|         | 20 52 | M | Right | 7.5 | Single | N/A |
|         | 21 69 | M | Left | 10 | Single | N/A |
| Imaizu et al\(^22\) | 22 77 | M | Left | 6 | Single | Cholangiocarcinoma |
| Honda et al\(^23\) | 23 61 | F | N/A | N/A | Multiple | IHCC |
| Itamoto et al\(^24\) | 24 70 | M | Right | 8 | Single | HCC |
| Matsuo et al\(^25\) | 25 77 | F | Left | 7.7 | Single | Hepatic abscess |
| Kaibori et al\(^1\) | 26 69 | F | Left | 22 | Single | Hepatic carcinoma |
| Lim et al\(^26\) | 27 41 | F | Left | 17 | Single | Hepatic mass |
| Sato et al\(^8\) | 28 87 | M | Left | 4 | Single | IHCC |
| Malhotra et al\(^2\) | 29 60 | F | Right | 20 | Single | Hepatic carcinoma |
| Bilgin et al\(^27\) | 30 48 | M | Left | 13 | Single | Hepatic carcinoma |
| Watanabe et al\(^12\) | 31 62 | M | Right | 5 | Multiple | IHCC |
| Gu et al\(^13\) | 32 65 | M | N/A | N/A | Single | N/A |
|         | 33 70 | M | N/A | N/A | Single | N/A |
|         | 34 48 | F | N/A | N/A | Single | N/A |
|         | 35 45 | M | N/A | N/A | Single | N/A |
|         | 36 46 | F | N/A | N/A | Single | N/A |
|         | 37 69 | M | N/A | N/A | Single | N/A |
|         | 38 54 | F | N/A | N/A | Single | N/A |
|         | 39 74 | M | N/A | N/A | Single | N/A |
|         | 40 57 | M | N/A | N/A | Single | N/A |
|         | 41 51 | M | N/A | N/A | Single | N/A |
|         | 42 69 | M | N/A | N/A | Single | N/A |
|         | 43 61 | F | N/A | N/A | Single | N/A |
|         | 44 53 | M | N/A | N/A | Single | N/A |
| Ning et al\(^4\) | 45 63 | M | Left | 8 | Multiple | Cholangiocarcinoma |
| Our case | 46 64 | M | Left | 2 | Single | Hepatic mass |

N/A: not available; S-iCCA: sarcomatoid intrahepatic cholangiocarcinoma; HCC: hepatocellular carcinoma; IHCC: intrahepatic cholangiocarcinoma; F: female; M: male.
| Case no. | AST (U/L) | ALT (U/L) | GGT (U/L) | CEA (ng/mL) | CA19-9 (U/mL) |
|---------|-----------|-----------|-----------|-------------|---------------|
| 1       | 25        | 19        | 115       | 0.74        | >1200.00      |
| 2       | 31        | 10        | 32        | 1.45        | 3.38          |
| 3       | 54        | 96        | 137       | 0.1         | 3             |
| 4       | 42        | 30        | N/A       | 2.35        | 1809.57       |
| 5       | 43        | 57        | 203       | 1.81        | 2.33          |
| 6       | 23        | 39        | 253       | 12.7        | 710.38        |
| 7       | 23        | 16        | 224       | 1.18        | 12.59         |
| 8       | 30        | 31        | 323       | 3.15        | >1200.00      |
| 9       | 80        | 30        | 98        | 1.08        | <2.00         |
| 10      | 37        | 47        | N/A       | 3.56        | 599.14        |
| 11      | 34        | 36        | 35        | 1.81        | 5.77          |
| 12      | 20        | 15        | N/A       | N/A         | normal        |
| 13      | 34        | N/A       | 77        | normal      | normal        |
| 14      | 75        | 46        | 356       | N/A         | N/A           |
| 15      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 16      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 17      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 18      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 19      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 20      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 21      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 22      | 33        | 27        | 99        | <0.5        | 17            |
| 23      | 22        | 30        | 175       | 9           | 13394         |
| 24      | normal    | 348       | normal    | normal      | normal        |
| 25      | N/A       | N/A       | N/A       | normal      | normal        |
| 26      | N/A       | N/A       | N/A       | normal      | normal        |
| 27      | N/A       | N/A       | N/A       | normal      | normal        |
| 28      | N/A       | N/A       | N/A       | 16.2        | 2894          |
| 29      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 30      | 152       | 45        | 297       | N/A         | 39            |
| 31      | 174       | 356       | 405       | 1.4         | 1109.9        |
| 32      | N/A       | N/A       | N/A       | N/A         | 11.25         |
| 33      | N/A       | N/A       | N/A       | N/A         | 22.44         |
| 34      | N/A       | N/A       | N/A       | N/A         | 7.28          |
| 35      | N/A       | N/A       | N/A       | N/A         | 10384         |
| 36      | N/A       | N/A       | N/A       | N/A         | 25.81         |
| 37      | N/A       | N/A       | N/A       | N/A         | 11.34         |
| 38      | N/A       | N/A       | N/A       | N/A         | 6.07          |
| 39      | N/A       | N/A       | N/A       | N/A         | 2             |
| 40      | N/A       | N/A       | N/A       | N/A         | 11.71         |
| 41      | N/A       | N/A       | N/A       | N/A         | N/A           |
| 42      | N/A       | N/A       | N/A       | N/A         | 886.51        |
| 43      | N/A       | N/A       | N/A       | N/A         | 10.55         |
| 44      | N/A       | N/A       | N/A       | N/A         | 2             |
| 45      | 25        | 19        | 125       | 2.17        | 100.5         |
| 46      | normal    | normal    | 119.3     | normal      | 351.74        |

AST: aspartate aminotransferase; ALT: alanine aminotransferase; GGT: gamma-glutamyl transferase; CEA: carcinoembryonic antigen; CA19-9: carbohydrate antigen 19-9; N/A: not available.
| Case no. | Positive results | Negative results |
|---------|------------------|------------------|
| 1       | CK19, vimentin    | HSA, CD10        |
| 2       | CK, vimentin, CEA, AFP | CK7, CK19, HSA, c-kit, CD117 |
| 3       | CK, CK19, vimentin | CK8, desmin, EMA, CEA, c-kit, S-100 |
| 4       | CK, CK8, CK19, vimentin, CEA, EMA | HSA, AFP, TTF-1 |
| 5       | CK, CK8, CK19, vimentin, SMA | HSA, CD5, CD68, HMW-CK |
| 6       | CK7, CK8, CK19, vimentin, CEA | HSA |
| 7       | CK7, CK8, CK19, vimentin, CD34 | HSA, CEA, HMW-CK |
| 8       | CK19, vimentin, CEA, p53 | CD31, CD34 |
| 9       | CK19, vimentin, CEA | CK7, desmin, HSA, SMA, c-kit, S-100 |
| 10      | CK, CK19, vimentin, CEA | HSA, CD31 |
| 11      | CK7, CK19, vimentin, MUC1 | HSA, CD10 |
| 12      | CK7, vimentin     | CK20, HepPar1 |
| 13      | KER, EMA, vimentin | AFP, S-100, AAT |
| 14      | low molecular cytokeratin, vimentin | UEA-1, desmin |
| 15      | KER, EMA, CA19-9 | PAS, CEA, AFP, vimentin, actin, desmin, S-100, NSE |
| 16      | KER, EMA, vimentin | PAS, CEA, AFP, CA199, actin, desmin, S-100, NSE |
| 17      | /                | PAS, CEA, AFP, CA199, actin, desmin, S-100, NSE, KER, EMA, vimentin |
| 18      | PAS, KER, EMA, vimentin | CEA, CA199, AFP, actin, desmin, S-100, NSE |
| 19      | KER, EMA         | PAS, CEA, AFP, CA199, actin, desmin, S-100, NSE, vimentin |
| 20      | PAS, KER, EMA, CEA | vimentin, CA199, AFP, actin, desmin, S-100, NSE |
| 21      | /                | PAS, CEA, AFP, CA199, actin, desmin, S-100, NSE, KER, EMA, vimentin |
| 22      | wide-spectrum keratin, vimentin, CEA | muscle actin, AAT, S-100, AFP |
| 23      | vimentin         | S-100, desmin, AFP, albumin, myoglobin |
| 24      | KER, EMA, vimentin | AFP, CEA, CA199, actin, desmin, S-100 |
| 25      | AAT, vimentin, F13a | desmin, EMA, CYT, SMA, CEA, AFP |
| 26      | vimentin, EMA, CK | S-100, CEA, AFP |
| 27      | CK-pan, vimentin, CEA | CK7, CK20, S-100, HMB-45, AMA, CD34, AFP, c-Kit |
| 28      | CK19, vimentin, CD44s | β-catenin |
| 29      | EMA, AE1/AE3, CK7, CK19, CEA | HepPar-1 |
| 30      | N/A              | N/A |
| 31      | CK, vimentin     | N/A |
| 32      | N/A              | N/A |
| 33      | N/A              | N/A |
| 34      | N/A              | N/A |
| 35      | N/A              | N/A |
| 36      | N/A              | N/A |
| 37      | N/A              | N/A |
| 38      | N/A              | N/A |
| 39      | N/A              | N/A |

(continued)
Table 3. Continued.

| Case no. | Positive results | Negative results |
|----------|------------------|------------------|
| 40       | N/A              | N/A              |
| 41       | N/A              | N/A              |
| 42       | N/A              | N/A              |
| 43       | N/A              | N/A              |
| 44       | N/A              | N/A              |
| 45       | AE1/AE3, STAT6, SOX10, CD34, CK19, desmin, MUC1, vimentin, SMA, S-100 | N/A |
| 46       | CK-pan, CK8, vimentin | CK7, CK20, HepPar-1 |

CA19-9: carbohydrate antigen 19-9; CEA: carcinoembryonic antigen; CD10: cluster of differentiation 10; NSE: neuron-specific enolase; AFP: a-fetoprotein; N/A: not available; PAS: periodic acid–Schiff; KER: keratin; EMA: epithelial membrane antigen; CK: cytokeratin; MUC1: mucin-1; HSA: human serum albumin; AAT: α-1-antitrypsin; SMA: smooth muscle actin; AMA: anti-mitochondria autoantibodies; UEA-1: ulex europaeus agglutinin-1; TTF-1: thyroid transcription factor-1; HMW-CK: high molecular weight cytokeratin; c-kit: receptor tyrosine kinase; CYT: cytochrome; HMB-45: human melanoma black 45; SOX-10: SRY-related HMG-BOX Gene 10; STAT-6: signal transducer and activator of transcription 6; F13a: factor XIIIa; AE1/AE3=CK-pan; HepPar-1: hepatocyte paraffin 1.

Table 4. Prognosis of S-iCCA patients reported in the English language literature.

| Case no. | Survival (months) | Treatment | Outcome |
|----------|-------------------|-----------|---------|
| 1        | 1.7               | Chemotherapy | Died    |
| 2        | 4.9               | Chemotherapy | Died    |
| 3        | 4.3               | Chemotherapy | Died    |
| 4        | 0.7               | Supportive   | Died    |
| 5        | 2.4               | Chemotherapy | Died    |
| 6        | 4.2               | Chemotherapy | Died    |
| 7        | 0.6               | Supportive   | Died    |
| 8        | 1.0               | Chemotherapy | Died    |
| 9        | 1.4               | Chemotherapy | N/A     |
| 10       | 0.5               | Supportive   | Died    |
| 11       | 12.8              | Viscum album | Alive   |
| 12       | 1.5               | Operation    | Died    |
| 13       | N/A               | None        | N/A     |
| 14       | 1.0               | None        | Died    |
| 15       | 3.0               | None        | Died    |
| 16       | 4.5               | Operation    | Died    |
| 17       | 5.0               | Chemotherapy | Died    |
| 18       | 2.5               | None        | Died    |
| 19       | 1.0               | TAE         | Died    |
| 20       | 2.0               | TAE         | Died    |
| 21       | 36.0              | Operation   | Alive   |
| 22       | 11.0              | Operation   | Alive   |
| 23       | 3.8               | None        | Died    |
| 24       | 9.0               | Operation   | Alive   |

(continued)
not undergo surgery was 3 months, which is considerably shorter than that of patients with ordinary iCCA.\textsuperscript{12,13} Several studies have shown that cisplatin, doxorubicin, cyclophosphamide, and taxol as adjuvant chemotherapy after surgery may prolong survival in patients with sarcomatoid carcinomas.\textsuperscript{14–17} In the patient described here, we learned by telephone follow-up that tumor metastasis and recurrence precluded further chemotherapy, and the patient died 3 months after surgery. To help improve the prognosis of patients with S-iCCA, early detection and radical surgery with careful follow-up are necessary, and this case demonstrates the importance of closer follow-up in patients with these malignancies. Furthermore, more comprehensive treatment options need to be explored.

In summary, S-iCCA is a rare malignancy. Diagnosis is only possible using pathology and immunohistochemical analyses because clinicomorphological findings and serologic and radiologic examinations are not disease-specific. Because of its high invasiveness, the prognosis of S-iCCA is poor, and closer follow-up is critical.

**Ethics statement**
Ethics permission was not obtained because our paper is a case report. The patient provided consent to publish this paper.

**Declaration of conflicting interest**
The authors declare that there is no conflict of interest.

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