

World Journal of *Clinical Cases*

World J Clin Cases 2022 September 6; 10(25): 8808-9179



Contents

Thrice Monthly Volume 10 Number 25 September 6, 2022

MINIREVIEWS

- 8808 Ear, nose, and throat manifestations of COVID-19 and its vaccines
Al-Ani RM
- 8816 Potential influences of religiosity and religious coping strategies on people with diabetes
Onyishi CN, Eseadi C, Ilechukwu LC, Okoro KN, Okolie CN, Egbule E, Asogwa E

ORIGINAL ARTICLE

Case Control Study

- 8827 Effectiveness of six-step complex decongestive therapy for treating upper limb lymphedema after breast cancer surgery
Zhang HZ, Zhong QL, Zhang HT, Luo QH, Tang HL, Zhang LJ

Retrospective Study

- 8837 Hospital admissions from alcohol-related acute pancreatitis during the COVID-19 pandemic: A single-centre study
Mak WK, Di Mauro D, Pearce E, Karran L, Myintmo A, Duckworth J, Orabi A, Lane R, Holloway S, Manzelli A, Mossadegh S
- 8844 Indocyanine green plasma clearance rate and 99mTc-galactosyl human serum albumin single-photon emission computed tomography evaluated preoperative remnant liver
Iwaki K, Kaihara S, Kita R, Kitamura K, Hashida H, Uryuhara K
- 8854 Arthroscopy with subscapularis upper one-third tenodesis for treatment of recurrent anterior shoulder instability independent of glenoid bone loss
An BJ, Wang FL, Wang YT, Zhao Z, Wang MX, Xing GY
- 8863 Evaluation of the prognostic nutritional index for the prognosis of Chinese patients with high/extremely high-risk prostate cancer after radical prostatectomy
Yang F, Pan M, Nie J, Xiao F, Zhang Y

Observational Study

- 8872 Chlorine poisoning caused by improper mixing of household disinfectants during the COVID-19 pandemic: Case series
Lin GD, Wu JY, Peng XB, Lu XX, Liu ZY, Pan ZG, Qiu ZW, Dong JG
- 8880 Mental health of the Slovak population during COVID-19 pandemic: A cross-sectional survey
Kralova M, Brazinova A, Sivcova V, Izakova L

Prospective Study

- 8893** Arthroscopic anatomical reconstruction of lateral collateral ligaments with ligament advanced reinforcement system artificial ligament for chronic ankle instability
Wang Y, Zhu JX

SYSTEMATIC REVIEWS

- 8906** How to select the quantitative magnetic resonance technique for subjects with fatty liver: A systematic review
Li YW, Jiao Y, Chen N, Gao Q, Chen YK, Zhang YF, Wen QP, Zhang ZM
- 8922** Lymphocytic choriomeningitis virus: An under-recognized congenital teratogen
Ferenc T, Vujica M, Mrzljak A, Vilibic-Cavlek T

CASE REPORT

- 8932** Alagille syndrome associated with total anomalous pulmonary venous connection and severe xanthomas: A case report
Zeng HS, Zhang ZH, Hu Y, Zheng GL, Wang J, Zhang JW, Guo YX
- 8939** Colo-colonic intussusception with post-polypectomy electrocoagulation syndrome: A case report
Moon JY, Lee MR, Yim SK, Ha GW
- 8945** Portal vein gas combined with pneumatosis intestinalis and emphysematous cystitis: A case report and literature review
Hu SF, Liu HB, Hao YY
- 8954** Quadricuspid aortic valve and right ventricular type of myocardial bridging in an asymptomatic middle-aged woman: A case report
Sopek Merkaš I, Lakušić N, Paar MH
- 8962** Treatment of gastric carcinoma with lymphoid stroma by immunotherapy: A case report
Cui YJ, Ren YY, Zhang HZ
- 8968** Gallstone associated celiac trunk thromboembolisms complicated with splenic infarction: A case report
Wu CY, Su CC, Huang HH, Wang YT, Wang CC
- 8974** Extracorporeal membrane oxygenation for lung cancer-related life-threatening hypoxia: A case report
Yoo SS, Lee SY, Choi SH
- 8980** Multi-disciplinary treatment of maxillofacial skeletal deformities by orthognathic surgery combined with periodontal phenotype modification: A case report
Liu JY, Li GF, Tang Y, Yan FH, Tan BC
- 8990** X-linked recessive Kallmann syndrome: A case report
Zhang P, Fu JY
- 8998** Delayed complications of intradural cement leakage after percutaneous vertebroplasty: A case report
Ma QH, Liu GP, Sun Q, Li JG

- 9004** Coexistent Kaposi sarcoma and post-transplant lymphoproliferative disorder in the same lymph nodes after pediatric liver transplantation: A case report
Zhang SH, Chen GY, Zhu ZJ, Wei L, Liu Y, Liu JY
- 9012** Misdiagnosis of pancreatic metastasis from renal cell carcinoma: A case report
Liang XK, Li LJ, He YM, Xu ZF
- 9020** Discoid medial meniscus of both knees: A case report
Zheng ZR, Ma H, Yang F, Yuan L, Wang GD, Zhao XW, Ma LF
- 9028** Simultaneous laparoscopic and arthroscopic excision of a huge juxta-articular ganglionic cyst compressing the sciatic nerve: A case report
Choi WK, Oh JS, Yoon SJ
- 9036** One-stage revision arthroplasty in a patient with ochronotic arthropathy accompanied by joint infection: A case report
Wang XC, Zhang XM, Cai WL, Li Z, Ma C, Liu YH, He QL, Yan TS, Cao XW
- 9044** Bladder paraganglioma after kidney transplantation: A case report
Wang L, Zhang YN, Chen GY
- 9050** Total spinal anesthesia caused by lidocaine during unilateral percutaneous vertebroplasty performed under local anesthesia: A case report
Wang YF, Bian ZY, Li XX, Hu YX, Jiang L
- 9057** Ruptured splenic artery aneurysms in pregnancy and usefulness of endovascular treatment in selective patients: A case report and review of literature
Lee SH, Yang S, Park I, Im YC, Kim GY
- 9064** Gastrointestinal metastasis secondary to invasive lobular carcinoma of the breast: A case report
Li LX, Zhang D, Ma F
- 9071** Post-bulbar duodenal ulcer with anterior perforation with kissing ulcer and duodenocaval fistula: A case report and review of literature
Alzerwi N
- 9078** Modified orthodontic treatment of substitution of canines by first premolars: A case report
Li FF, Li M, Li M, Yang X
- 9087** Renal cell carcinoma presented with a rare case of icteric Stauffer syndrome: A case report
Popov DR, Antonov KA, Atanasova EG, Pentchev CP, Milatchkov LM, Petkova MD, Neykov KG, Nikolov RK
- 9096** Successful resection of a huge retroperitoneal venous hemangioma: A case report
Qin Y, Qiao P, Guan X, Zeng S, Hu XP, Wang B
- 9104** Malignant transformation of biliary adenofibroma combined with benign lymphadenopathy mimicking advanced liver carcinoma: A case report
Wang SC, Chen YY, Cheng F, Wang HY, Wu FS, Teng LS

- 9112** Congenital hepatic cyst: Eleven case reports
Du CX, Lu CG, Li W, Tang WB
- 9121** Endovascular treatment of a ruptured pseudoaneurysm of the internal carotid artery in a patient with nasopharyngeal cancer: A case report
Park JS, Jang HG
- 9127** Varicella-zoster virus meningitis after spinal anesthesia: A case report
Lee YW, Yoo B, Lim YH
- 9132** Chondrosarcoma of the toe: A case report and literature review
Zhou LB, Zhang HC, Dong ZG, Wang CC
- 9142** Tamsulosin-induced life-threatening hypotension in a patient with spinal cord injury: A case report
Lee JY, Lee HS, Park SB, Lee KH
- 9148** CCNO mutation as a cause of primary ciliary dyskinesia: A case report
Zhang YY, Lou Y, Yan H, Tang H
- 9156** Repeated bacteremia and hepatic cyst infection lasting 3 years following pancreatoduodenectomy: A case report
Zhang K, Zhang HL, Guo JQ, Tu CY, Lv XL, Zhu JD
- 9162** Idiopathic cholesterol crystal embolism with atheroembolic renal disease and blue toes syndrome: A case report
Cheng DJ, Li L, Zheng XY, Tang SF
- 9168** Systemic lupus erythematosus with visceral varicella: A case report
Zhao J, Tian M

LETTER TO THE EDITOR

- 9176** Imaging of fibroadenoma: Be careful with imaging follow-up
Ece B, Aydın S

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Mohsen Khosravi, MD, Assistant Professor, Department of Psychiatry and Clinical Psychology, Zahedan University of Medical Sciences, Zahedan 9819713955, Iran. m.khosravi@zaums.ac.ir

AIMS AND SCOPE

The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The WJCC is now abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Journal Citation Reports/Science Edition, Current Contents®/Clinical Medicine, PubMed, PubMed Central, Scopus, Reference Citation Analysis, China National Knowledge Infrastructure, China Science and Technology Journal Database, and Superstar Journals Database. The 2022 Edition of Journal Citation Reports® cites the 2021 impact factor (IF) for WJCC as 1.534; IF without journal self cites: 1.491; 5-year IF: 1.599; Journal Citation Indicator: 0.28; Ranking: 135 among 172 journals in medicine, general and internal; and Quartile category: Q4. The WJCC's CiteScore for 2021 is 1.2 and Scopus CiteScore rank 2021: General Medicine is 443/826.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Xu Guo*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei Wang*.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

September 6, 2022

COPYRIGHT

© 2022 Baishideng Publishing Group Inc

INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>



Malignant transformation of biliary adenofibroma combined with benign lymphadenopathy mimicking advanced liver carcinoma: A case report

Shao-Cheng Wang, Yan-Yan Chen, Fei Cheng, Hai-Yong Wang, Fu-Sheng Wu, Li-Song Teng

Specialty type: Oncology

Provenance and peer review:

Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

P-Reviewer: Ahmed SK, Iraq; Trébol J, Spain

Received: April 14, 2022

Peer-review started: April 14, 2022

First decision: May 30, 2022

Revised: June 12, 2022

Accepted: July 20, 2022

Article in press: July 20, 2022

Published online: September 6, 2022



Shao-Cheng Wang, Yan-Yan Chen, Hai-Yong Wang, Fu-Sheng Wu, Li-Song Teng, Department of Surgical Oncology, The First Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310000, Zhejiang Province, China

Fei Cheng, Department of Pathology, The First Affiliated Hospital, Zhejiang University School of Medicine, Hangzhou 310000, Zhejiang Province, China

Corresponding author: Li-Song Teng, PhD, Professor, Department of Surgical Oncology, First Affiliated Hospital, Zhejiang University School of Medicine, No. 79 Qingchun Road, Hangzhou 310000, Zhejiang Province, China. lsteng@zju.edu.cn

Abstract

BACKGROUND

Biliary adenofibromas (BAFs) are rare primary hepatic neoplasms, some of which can potentially undergo malignant transformation. Here, we describe a rare case of malignant transformation of BAF.

CASE SUMMARY

A 51-year-old female was referred to our hospital with epigastric pain. Computed tomography showed a solitary liver mass combined with the enlargement of multiple mediastinal and cervical lymph nodes, clinically mimicking a liver carcinoma with extensive lymph node metastasis. However, core needle biopsy suggested BAF with malignant transformation. Finally, the patient underwent curative resection of the neoplasm and was recurrence-free for 12 mo.

CONCLUSION

Our case serves as an example of a rare manifestation of BAF. Our report and the previously published experience, reinforce that curative resection should be considered the primary treatment for BAFs with malignant transformation, leading to a favorable prognosis.

Key Words: Biliary adenofibroma; Malignant transformation; Lymphadenopathy; Surgery; Case report

©The Author(s) 2022. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: Biliary adenofibromas (BAFs) are rare primary hepatic neoplasms, some of which have potential of malignant transformation. Here we describe a rare case of malignant transformation of BAF presented with epigastric pain, whose imaging showed solitary liver mass combined with enlargement of multiple mediastinal and cervical lymph nodes, clinically mimicking a liver carcinoma with extensive lymph node metastasis. The patient was treated with curative resection of the neoplasm and has been recurrence-free for 12 mo. Our case is a rare manifestation of BAF and our experience supported that curative resection should be considered the primary treatment for BAFs with malignant transformation, which leads to a favorable prognosis.

Citation: Wang SC, Chen YY, Cheng F, Wang HY, Wu FS, Teng LS. Malignant transformation of biliary adenofibroma combined with benign lymphadenopathy mimicking advanced liver carcinoma: A case report. *World J Clin Cases* 2022; 10(25): 9104-9111

URL: <https://www.wjgnet.com/2307-8960/full/v10/i25/9104.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v10.i25.9104>

INTRODUCTION

Biliary adenofibromas (BAFs) are rare primary neoplasms of the liver first described by Tsui *et al*[1] in 1993, most of which are benign biliary cystic tumors[2,3]. Although BAFs are benign tumor with indolent biological behavior, some BAFs have the potential to undergo malignant transformation, leading to transition to invasive carcinoma. To date, few cases of BAFs with malignant features have been reported[4,5].

In the present report, we describe an extremely rare case of malignant transformation of BAF combined with benign lymphadenopathy, which clinically mimicked liver carcinoma with extensive lymph node metastasis. The patient underwent a complete surgical resection of the disease. Meanwhile, we provide a brief review of literature concerning malignant transformation of BAFs.

CASE PRESENTATION

Chief complaints

A 51-year-old female with a 2-mo history of epigastric pain.

History of present illness

The pain was barely noticeable initially and steadily worsened thereafter. On the initial evaluation, she had loss of appetite, a 5 kg weight loss, and denied nausea, vomiting, or jaundice. The patient had periodic episodes of fever ranging from 37.5°C to 38.6°C for about a week.

History of past illness

The patient had laparoscopic cholecystectomy 6 years ago. Other medical, surgical, psychosocial, and family histories were non-contributory.

Personal and family history

The patient denied any family history.

Physical examination

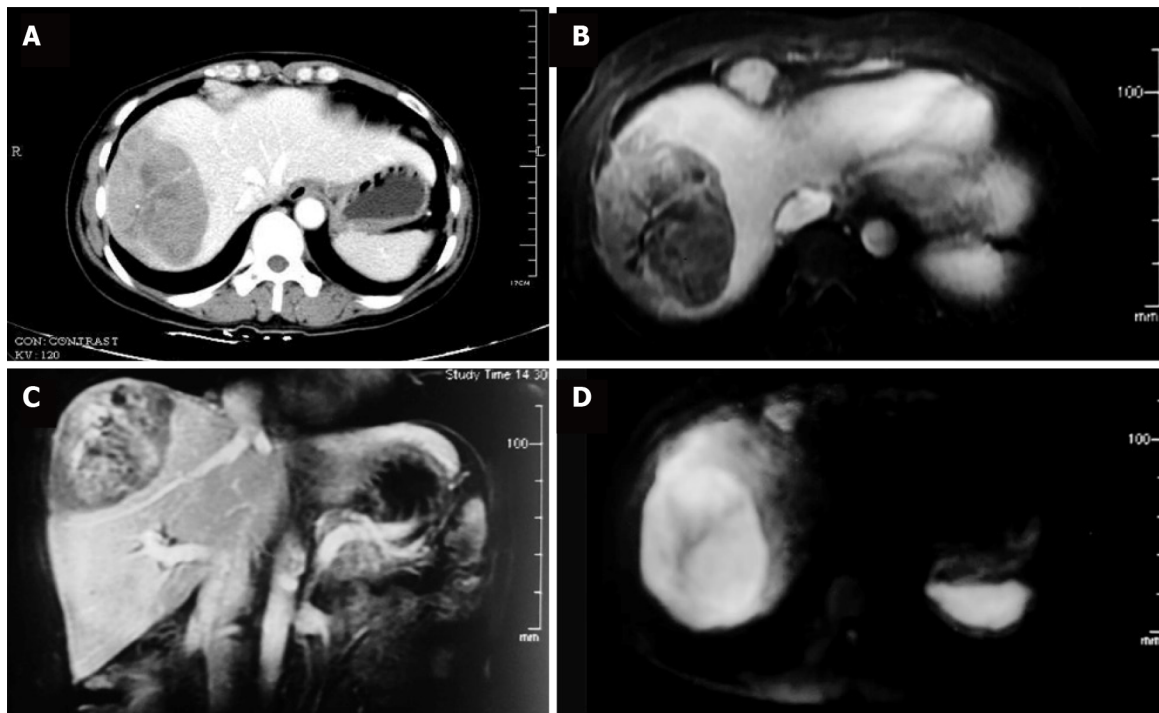
Physical examination revealed mild epigastric tenderness that was otherwise unremarkable.

Laboratory examinations

The patient had an elevated C-reactive protein (CRP) of 75.1 mg/L, while the white blood cell count was within the normal range. The tumor markers alpha-fetoprotein, carcinoembryonic antigen, and cancer antigen 19-9 were within normal ranges. Liver function was normal, and serology was negative for hepatitis B and hepatitis C virus infections.

Imaging examinations

The patient was referred to an oncologic surgeon. Contrast abdominal computed tomography (CT), and subsequent magnetic resonance imaging showed a 9.2 cm × 5.9 cm solitary mass in hepatic segments VII and VIII, which was unevenly enhanced during the arterial phase and washed out during the venous phase (Figure 1). Additionally, pulmonary CT showed multiple enlarged merging mediastinal and



DOI: 10.12998/wjcc.v10.i25.9104 Copyright ©The Author(s) 2022.

Figure 1 Computed tomography, magnetic resonance imaging and positron emission tomography/computed tomography images of liver mass and mediastinal lymphadenopathy. A: Abdominal contrast computed tomography, venous phase; B-D: Representative images from the MRI study (B: Venous phase; C: Sagittal venous phase; D: Diffusion weighted).

cervical lymph nodes (Figure 2A). The leading diagnosis based on imaging was malignant liver carcinoma, possibly hepatocellular carcinoma or intrahepatic cholangiocarcinoma, with extensive lymph node metastasis. To exclude possible distant metastasis, positron emission tomography/computed tomography was conducted, which showed uneven fluorodeoxyglucose (FDG) uptake (maximum SUV 6.7) in the liver mass, elevated FDG uptake (maximum SUV 1.9) in multiple mediastinal and cervical lymph nodes (Figure 2B and C), and no suspicious FDG elevation in the lung, brain, or pelvic cavity.

FINAL DIAGNOSIS

A core needle biopsy was suggested to guide systemic treatment. Unexpectedly, the pathological finding for the biopsy suggested an epithelial neoplasm with a tubuloglandular structure. The epithelial cells immunohistochemically stained positive for cytokeratin (CK) 7 and CK19 but negative for glypican-3, p53, and hepatocyte-specific antigen, implying a biliary origin. The ki-67 index was approximately 5%. The morphology and molecular pathology were compatible with adenofibroma with a malignant transformation. The patient underwent a cervical lymph node excisional biopsy to exclude distant lymph node metastasis. Three swollen lymph nodes were resected, which were negative for cancerous components.

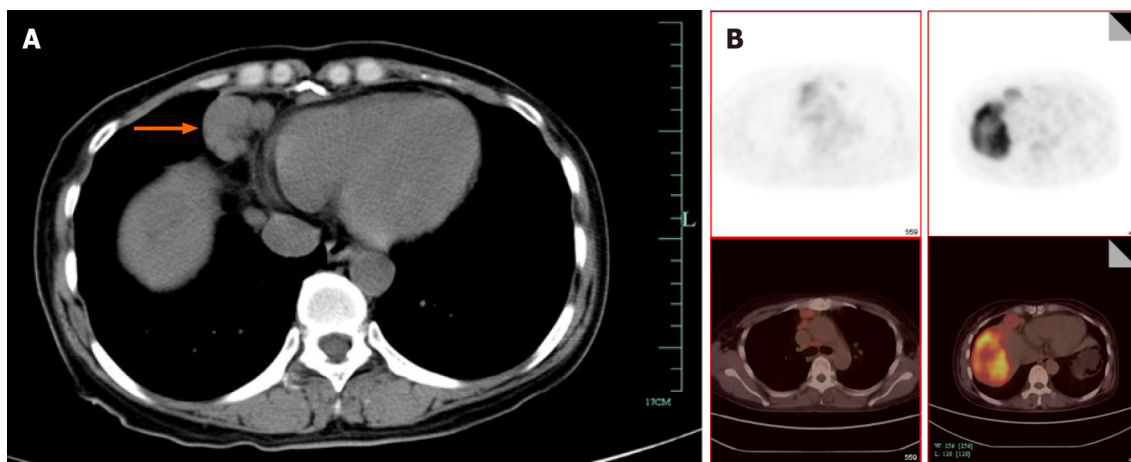
TREATMENT

Our team decided to perform surgery on this patient based on the current knowledge of biliary adenofibroma with malignant transformation. Complete local resection of the hepatic lesion and resection of multiple merged lymph nodes in the anterior-inferior mediastinum were performed (the macroscopic view is presented in Figure 3). In detail, under general anesthesia, open atypical hepatectomy was done by Cavitron Ultrasonic Surgical Aspirator to remove the tumor with a margin of at least 1cm. The mediastinum lymph nodes were dissected through the trans-esophageal hiatus approach. The surgery lasted 3.5 h and estimated blood lost was 200 mL. The patient was well-recovered and discharged 11 days after surgery (Table 1). Subsequent pathological findings of the hepatic lesion were consistent with those of needle biopsy. The resected tissue was composed of epithelial cells with irregular tubular shapes, exhibiting cytological atypia and collagenous stroma with

Table 1 Timeline of management

Time (day)	Event
1	Outpatient evaluation
4	Hospitalization
6	Found liver mass by CT
11	PET-CT
13	Core needle biopsy of the liver mass
15	Cervical lymph node biopsy
25	Surgery
36	Discharge

CT: Computed tomography; PET: Positron emission tomography.



DOI: 10.12998/wjcc.v10.i25.9104 Copyright ©The Author(s) 2022.

Figure 2 computed tomography images. A: Pulmonary computed tomography, mediastinal window; B: Representative positron emission tomography/computed tomography images, left, mediastinum, right, abdominal.

plasmocytic infiltration (Figure 4A and B). All margins were negative. Again, no neoplastic composition was found in the resected lymph nodes, whereas lymphatic hyperplasia with massive plasmocytic infiltration was observed.

Immunohistochemical staining was positive for CD3, CD20, CD10, CD21, KappaK, Lambda, and IgG4 (Figure 4C) but negative for HHV8 (Figure 4D). For the differential diagnosis of immune-mediated diseases such as IgG4-related lymphadenopathy, the patient's serum IgG4, IgE, and IL-6 levels were tested, all within normal ranges.

OUTCOME AND FOLLOW-UP

The patient did not develop recurrence of the hepatic lesion during the 12 mo postoperative follow-up (assessed by local CT); no fever, epigastric pain, or other discomfort was present.

DISCUSSION

BAF is a rare primary hepatic tumor first described in 1993 by Tsui *et al*[1]. It is pathologically characterized by dilated ductular structures lined with bland cuboidal epithelial cells and an abundant fibroblastic stroma. BAFs are generally regarded as benign lesions, but they should be considered premalignant neoplasms because of their potential for malignant transformation[2,6]. The literature has limited reports of BAFs with malignant features, with only 13 cases to date (Table 2). Based on a recent review[7], the malignant transformation was found in 37% (7/19) of BAF cases that underwent

Table 2 Biliary adenofibroma with malignant transformation of the liver, review of the literature

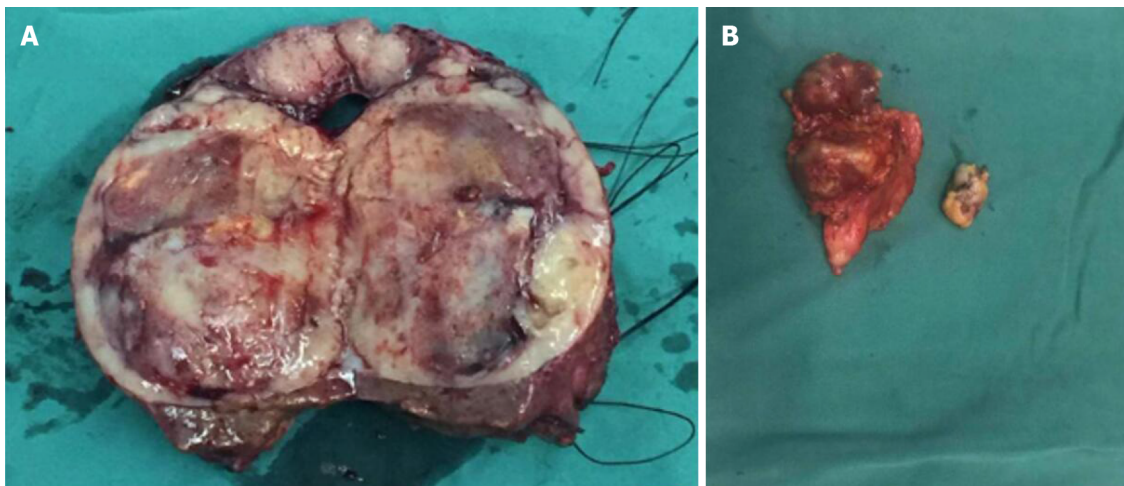
No.	Ref.	Age	Sex	Tumor location	Size (cm)	Treatment	Ki67, %	p53	Outcome	Additional information
1	Akin <i>et al</i> [8], 2002	25	M	Right hepatic lobe	20	Right hepatectomy	NA	NA	Hepatic recurrence and pulmonary metastasis after 3 yr	
2	Kai <i>et al</i> [12], 2012	40	M	Right hepatic lobe	7	Right hepatectomy	5-10	NA	No recurrence at 8-mo follow-up	
3	Nguyen <i>et al</i> [13], 2012	53	F	Segment IVb	6.5	Resection of segments III/IV	NA	NA	No recurrence after 12 mo	
4	Tsutsui <i>et al</i> [5], 2014	69	F	Segment VI	3.5	Partial hepatectomy	10-15	Focally positive	No recurrence at 4-yr follow-up	Dysplastic changes
5	Thompson <i>et al</i> [4], 2016	71	M	Left hepatic lobe	14.5	Left hepatectomy	NA	NA	No recurrence of liver tumor for 9 yr	Moderately-differentiated adenocarcinoma
6	Thompson <i>et al</i> [4], 2016	71	M	Caudate lobe	6.3	Caudate lobectomy	NA	NA	No recurrence at 1-mo follow-up	Well-differentiated adenocarcinoma
7	Godambe <i>et al</i> [6], 2016	71	F	Segments II, III and IVa	5.7	Left hepatectomy	50	Positive	NA	Microinvasive carcinoma
8	Thai <i>et al</i> [14], 2016	77	M	Segment II	4	Left hepatectomy	NA	NA	NA	Cholangiocarcinoma arising from BAF
9	Kaminsky <i>et al</i> [2], 2017	37	F	Segment V	4.5	Partial hepatectomy	50	Negative	No recurrence at 4-mo follow-up	Cholangiocarcinoma arising from BAF
10	Chua <i>et al</i> [7], 2018	66	F	Segment IVb	6	Wedge resection	2	Positive	No recurrence at 4-mo follow-up	Cholangiocarcinoma arising from BAF
11	Sturm <i>et al</i> [15], 2019	63	F	Segment IVa	6.3	Left hepatectomy	20-30	Focally positive	No recurrence at 24-mo follow-up	
12	Alshbib <i>et al</i> [9], 2022	63	M	Segments IVb and V	15.5	Resection of segments IVb and V	25	Weak	Hepatic recurrence 3 mo	
13	Current report	51	F	Segments VII and VIII	7.5	Partial hepatectomy	5	Negative	No recurrence at 12-mo follow-up	

BAF: Biliary adenofibroma; NA: Not available.

resection. Three cases were reported as cholangiocarcinoma arising from BAFs, 2 cases as adenocarcinomas, and 1 unspecified carcinoma, while the others contained some microscopic malignant features such as dysplastic changes and microinvasion. BAF has been reported to arise from liver cirrhosis secondary to chronic hepatitis C virus infection[7]. Immunohistochemistry (IHC) revealed that the epithelial component of the BAF was positive for CK7 and CK19. Of the 5 cases, 3 were positive for p53 on IHC staining. The Ki-67 index ranged from 2% to 50% in 6 cases with available data. Wide local resection is recommended as the primary treatment for BAFs with malignant transformation, and follow-up imaging is needed for potential recurrence[2]. Only 2 of the reported patients developed recurrence after surgery, presenting as local hepatic recurrence, and 1 with pulmonary metastasis[8,9], whereas other cases, with follow-up data from 1 mo to 9 years, had no postoperative recurrence. Therefore, despite the heterogeneity of the pathological features in these cases, BAFs with malignant transformation generally have a good prognosis after complete resection.

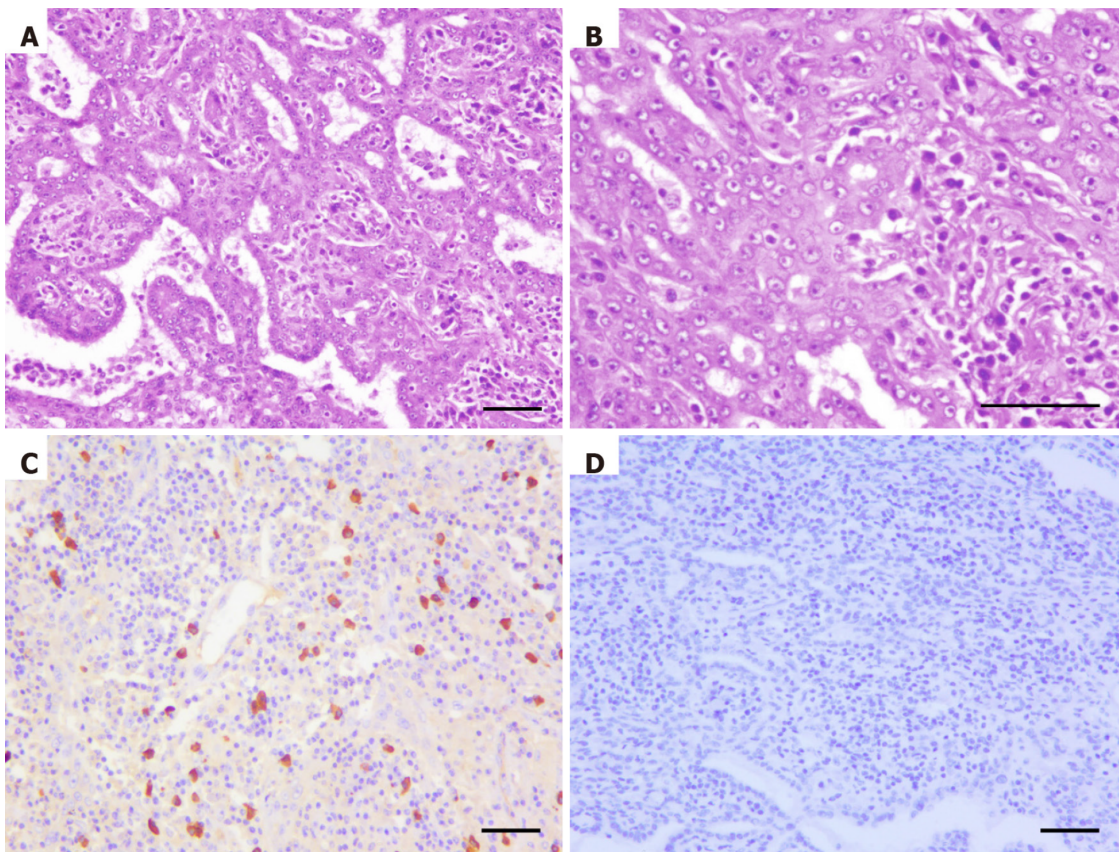
In the current case, the multiple enlarged lymph nodes were first misinterpreted as metastases arising from the hepatic neoplasm, leading to an initial diagnosis of advanced liver malignancy and, correspondingly, a plan for systemic treatment such as chemotherapy or targeted therapy. For advanced disease with high suspicion of hepatocellular carcinoma, pathology is not mandatory prior to systemic treatment. In this case, the imaging studies did not suggest a definite diagnosis, therefore, a core needle biopsy was conducted after prudent evaluation. Finally, the pathological diagnosis was BAF with malignant transformation, thus providing a rationale for complete resection of the neoplasm.

Lymphadenopathy showed no evidence of malignancy. It was first suspected to be IgG4-related or multicentric Castleman's disease due to its histology and positive IHC staining for IgG4[10,11]. However, the patient's serum IgG4, IgE, and IL-6 levels were within the normal range; therefore, we did not reach a definite diagnosis for her lymphadenopathy. The patient's fever was non-specific, which could be interpreted as an infection or neoplastic fever, apart from immune-mediated diseases. Notably, the fever did not recur, and CRP and erythrocyte sedimentation rates were normal after surgery.



DOI: 10.12998/wjcc.v10.i25.9104 Copyright ©The Author(s) 2022.

Figure 3 Macroscopic view of surgical specimens. A: Sectional appearance of the hepatic lesion; B: Enlarged mediastinal lymph node (multiple nodes merged). Bar = 1 cm.



DOI: 10.12998/wjcc.v10.i25.9104 Copyright ©The Author(s) 2022.

Figure 4 Microscopic pathology of surgical specimens. A and B: H & E staining of hepatic lesion, (A) H & E $\times 100$ (B) H & E $\times 200$. The malignant component of a tumor consists of deformed fused glandular ducts that form a sieve, and cord-like structures. The neoplastic cells are of medium size with well-defined nucleoli, and most of the cytoplasm is pale, slightly acidophilic or vacuolated; C and D: Immunohistochemical staining of IgG4 and HHV8 in mediastinal lymph node ($\times 100$). Bar = 100 μ m.

The limitation of this case is that, despite exclusive diagnosis for IgG4-related or multicentric Castleman's disease, we eventually did not reach a diagnosis for the patient's lymphadenopathy. Besides, we did not conduct molecular analysis such as MSI state and PD-L1 expression of the BAF sample. Additional molecular analysis may assist follow-up treatment of this disease in case of recurrence.

CONCLUSION

In summary, BAFs with malignant transformation are clinically unusual and could be misinterpreted as liver carcinoma based on imaging features, especially when comorbid with other suspicious signs of malignancy, such as lymphadenopathy. Our case serves as an example of a rare manifestation of BAF, a biopsy of suspicious lesions is vital for diagnosis. According to previous reports, BAFs with malignant transformation have the potential to develop into intrahepatic cholangiocarcinoma or adenocarcinoma, curative resection should be considered an essential primary treatment, which generally leads to a favorable outcome.

FOOTNOTES

Author contributions: Wang SC and Chen YY collected, analyzed the patient data and wrote the manuscript; Cheng F conducted the pathological analysis; Teng LS, Wang SC and Wang HY performed the operation and were in charge of the patient; Teng LS and Wu FS reviewed the manuscript; all authors have read and approve the final manuscript.

Supported by Innovative Project of Medical and Health Science, Zhejiang Province, No. 2021RC002.

Informed consent statement: The patient's informed consent has been obtained in this case.

Conflict-of-interest statement: All the authors have no financial disclosures or conflicts of interest to declare.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <https://creativecommons.org/licenses/by-nc/4.0/>

Country/Territory of origin: China

ORCID number: Fei Cheng 0000-0003-0557-3951; Li-Song Teng 0000-0001-6470-9017.

S-Editor: Liu JH

L-Editor: A

P-Editor: Liu JH

REFERENCES

- 1 Tsui WM, Loo KT, Chow LT, Tse CC. Biliary adenofibroma. A heretofore unrecognized benign biliary tumor of the liver. *Am J Surg Pathol* 1993; **17**: 186-192 [PMID: 8422113]
- 2 Kaminsky P, Preiss J, Sasatomi E, Gerber DA. Biliary adenofibroma: A rare hepatic lesion with malignant features. *Hepatology* 2017; **65**: 380-383 [PMID: 27631648 DOI: 10.1002/hep.28818]
- 3 Arnason T, Borger DR, Corless C, Hagen C, Iafrate AJ, Makhoul H, Misdraji J, Sapp H, Tsui WM, Wanless IR, Zuluaga Toro T, Lauwers GY. Biliary Adenofibroma of Liver: Morphology, Tumor Genetics, and Outcomes in 6 Cases. *Am J Surg Pathol* 2017; **41**: 499-505 [PMID: 28266931 DOI: 10.1097/PAS.0000000000000773]
- 4 Thompson SM, Zendejas-Mummert B, Hartgers ML, Venkatesh SK, Smyrk TC, Mahipal A, Smoot RL. Malignant transformation of biliary adenofibroma: a rare biliary cystic tumor. *J Gastrointest Oncol* 2016; **7**: E107-E112 [PMID: 28078134 DOI: 10.21037/jgo.2016.09.14]
- 5 Tsutsui A, Bando Y, Sato Y, Miyake H, Sawada-Kitamura S, Shibata H, Kakuda Y, Harada K, Sasaki M, Nakanuma Y. Biliary adenofibroma with ominous features of imminent malignant changes. *Clin J Gastroenterol* 2014; **7**: 441-448 [PMID: 26184026 DOI: 10.1007/s12328-014-0523-1]
- 6 Godambe A, Brunt EM, Fulling KH, Reza Kermanshahi T. Biliary Adenofibroma with Invasive Carcinoma: Case Report and Review of the Literature. *Case Rep Pathol* 2016; **2016**: 8068513 [PMID: 26885426 DOI: 10.1155/2016/8068513]
- 7 Chua D, Chiow AKH, Ang TL, Wang LM. Malignant Transformation Arising Within Unusual and Rare Hepatic Lesions: Fibropolycystic Disease Form of Ductal Plate Malformation and Biliary Adenofibroma. *Int J Surg Pathol* 2018; **26**: 542-550 [PMID: 29464972 DOI: 10.1177/1066896918758172]
- 8 Akin O, Coskun M. Biliary adenofibroma with malignant transformation and pulmonary metastases: CT findings. *AJR Am J Roentgenol* 2002; **179**: 280-281 [PMID: 12076957 DOI: 10.2214/ajr.179.1.1790280]
- 9 Alshbib A, Grzyb K, Syversveen T, Reims HM, Lassen K, Yaqub S. Biliary Adenofibroma: A Rare Liver Tumor with Transition to Invasive Carcinoma. *Case Rep Surg* 2022; **2022**: 5280884 [PMID: 35178267 DOI: 10.1155/2022/5280884]
- 10 Zhang X, Zhang P, Peng L, Fei Y, Zhang W, Feng R. Clinical characteristics of a concurrent condition of IgG4-RD and

- Castleman's disease. *Clin Rheumatol* 2018; **37**: 3387-3395 [PMID: [29948354](#) DOI: [10.1007/s10067-018-4165-4](#)]
- 11 **Stone JH**, Zen Y, Deshpande V. IgG4-related disease. *N Engl J Med* 2012; **366**: 539-551 [PMID: [22316447](#) DOI: [10.1056/NEJMr1104650](#)]
 - 12 **Kai K**, Yakabe T, Kohya N, Miyoshi A, Iwane S, Mizuta T, Miyazaki K, Tokunaga O. A case of unclassified multicystic biliary tumor with biliary adenofibroma features. *Pathol Int* 2012; **62**: 506-510 [PMID: [22726072](#) DOI: [10.1111/j.1440-1827.2012.02830.x](#)]
 - 13 **Nguyen NT**, Harring TR, Holley L, Goss JA, O'Mahony CA. Biliary adenofibroma with carcinoma in situ: a rare case report. *Case Reports Hepatol* 2012; **2012**: 793963 [PMID: [25374710](#) DOI: [10.1155/2012/793963](#)]
 - 14 **Thai E**, Dalla Valle R, Evaristi F, Silini EM. A case of biliary adenofibroma with malignant transformation. *Pathol Res Pract* 2016; **212**: 468-470 [PMID: [26778388](#) DOI: [10.1016/j.prp.2015.12.015](#)]
 - 15 **Sturm AK**, Welsch T, Meissner C, Aust DE, Baretton G. A case of biliary adenofibroma of the liver with malignant transformation: a morphomolecular case report and review of the literature. *Surg Case Rep* 2019; **5**: 104 [PMID: [31236706](#) DOI: [10.1186/s40792-019-0661-2](#)]



Published by **Baishideng Publishing Group Inc**
7041 Koll Center Parkway, Suite 160, Pleasanton, CA 94566, USA

Telephone: +1-925-3991568

E-mail: bpgoffice@wjgnet.com

Help Desk: <https://www.f6publishing.com/helpdesk>

<https://www.wjgnet.com>

