

World Journal of *Clinical Cases*

World J Clin Cases 2020 October 26; 8(20): 4688-5069



MINIREVIEWS

- 4688 Relationship between non-alcoholic fatty liver disease and coronary heart disease
Arslan U, Yenercağ M

ORIGINAL ARTICLE

Retrospective Cohort Study

- 4700 Remission of hepatotoxicity in chronic pulmonary aspergillosis patients after lowering trough concentration of voriconazole
Teng GJ, Bai XR, Zhang L, Liu HJ, Nie XH

Retrospective Study

- 4708 Endoscopic submucosal dissection as alternative to surgery for complicated gastric heterotopic pancreas
Noh JH, Kim DH, Kim SW, Park YS, Na HK, Ahn JY, Jung KW, Lee JH, Choi KD, Song HJ, Lee GH, Jung HY
- 4719 Observation of the effects of three methods for reducing perineal swelling in children with developmental hip dislocation
Wang L, Wang N, He M, Liu H, Wang XQ
- 4726 Predictive value of serum cystatin C for risk of mortality in severe and critically ill patients with COVID-19
Li Y, Yang S, Peng D, Zhu HM, Li BY, Yang X, Sun XL, Zhang M
- 4735 Sleep quality of patients with postoperative glioma at home
Huang Y, Jiang ZJ, Deng J, Qi YJ
- 4743 Early complications of preoperative external traction fixation in the staged treatment of tibial fractures: A series of 402 cases
Yang JZ, Zhu WB, Li LB, Dong QR
- 4753 Retroperitoneal vs transperitoneal laparoscopic lithotripsy of 20-40 mm renal stones within horseshoe kidneys
Chen X, Wang Y, Gao L, Song J, Wang JY, Wang DD, Ma JX, Zhang ZQ, Bi LK, Xie DD, Yu DX
- 4763 Undifferentiated embryonal sarcoma of the liver: Clinical characteristics and outcomes
Zhang C, Jia CJ, Xu C, Sheng QJ, Dou XG, Ding Y
- 4773 Cerebral infarct secondary to traumatic internal carotid artery dissection
Wang GM, Xue H, Guo ZJ, Yu JL
- 4785 Home-based nursing for improvement of quality of life and depression in patients with postpartum depression
Zhuang CY, Lin SY, Cheng CJ, Chen XJ, Shi HL, Sun H, Zhang HY, Fu MA

Observational Study

- 4793** Cost-effectiveness of lutetium (^{177}Lu) oxodotreotide *vs* everolimus in gastroenteropancreatic neuroendocrine tumors in Norway and Sweden
Palmer J, Leeuwenkamp OR
- 4807** Factors related to improved American Spinal Injury Association grade of acute traumatic spinal cord injury
Tian C, Lv Y, Li S, Wang DD, Bai Y, Zhou F, Ma QB
- 4816** Intraoperative systemic vascular resistance is associated with postoperative nausea and vomiting after laparoscopic hysterectomy
Qu MD, Zhang MY, Wang GM, Wang Z, Wang X

META-ANALYSIS

- 4826** Underwater *vs* conventional endoscopic mucosal resection in treatment of colorectal polyps: A meta-analysis
Ni DQ, Lu YP, Liu XQ, Gao LY, Huang X

CASE REPORT

- 4838** Dehydrated patient without clinically evident cause: A case report
Palladino F, Fedele MC, Casertano M, Liguori L, Esposito T, Guarino S, Miraglia del Giudice E, Marzuillo P
- 4844** Intracranial malignant solitary fibrous tumor metastasized to the chest wall: A case report and review of literature
Usuda D, Yamada S, Izumida T, Sangen R, Higashikawa T, Nakagawa K, Iguchi M, Kasamaki Y
- 4853** End-of-life home care of an interstitial pneumonia patient supported by high-flow nasal cannula therapy: A case report
Goda K, Kenzaka T, Kuriyama K, Hoshijima M, Akita H
- 4858** Rupture of carotid artery pseudoaneurysm in the modern era of definitive chemoradiation for head and neck cancer: Two case reports
Kim M, Hong JH, Park SK, Kim SJ, Lee JH, Byun J, Ko YH
- 4866** Unremitting diarrhoea in a girl diagnosed anti-N-methyl-D-aspartate-receptor encephalitis: A case report
Onpoaree N, Veeravigrom M, Sanpavat A, Suratannon N, Sintusek P
- 4876** Paliperidone palmitate-induced facial angioedema: A case report
Srifuengfung M, Sukakul T, Liangcheep C, Viravan N
- 4883** Improvement of lenvatinib-induced nephrotic syndrome after adaptation to sorafenib in thyroid cancer: A case report
Yang CH, Chen KT, Lin YS, Hsu CY, Ou YC, Tung MC
- 4895** Adult metaplastic hutch diverticulum with robotic-assisted diverticulectomy and reconstruction: A case report
Yang CH, Lin YS, Ou YC, Weng WC, Huang LH, Lu CH, Hsu CY, Tung MC

- 4902** Thrombus straddling a patent foramen ovale and pulmonary embolism: A case report
Huang YX, Chen Y, Cao Y, Qiu YG, Zheng JY, Li TC
- 4908** Therapeutic experience of an 89-year-old high-risk patient with incarcerated cholecystolithiasis: A case report and literature review
Zhang ZM, Zhang C, Liu Z, Liu LM, Zhu MW, Zhao Y, Wan BJ, Deng H, Yang HY, Liao JH, Zhu HY, Wen X, Liu LL, Wang M, Ma XT, Zhang MM, Liu JJ, Liu TT, Huang NN, Yuan PY, Gao YJ, Zhao J, Guo XA, Liao F, Li FY, Wang XT, Yuan RJ, Wu F
- 4917** Woven coronary artery: A case report
Wei W, Zhang Q, Gao LM
- 4922** Idiopathic multicentric Castleman disease with pulmonary and cutaneous lesions treated with tocilizumab: A case report
Han PY, Chi HH, Su YT
- 4930** Perianorectal abscesses and fistula due to ingested jujube pit in infant: Two case reports
Liu YH, Lv ZB, Liu JB, Sheng QF
- 4938** Forniceal deep brain stimulation in severe Alzheimer's disease: A case report
Lin W, Bao WQ, Ge JJ, Yang LK, Ling ZP, Xu X, Jiang JH, Zuo CT, Wang YH
- 4946** Systemic autoimmune abnormalities complicated by cytomegalovirus-induced hemophagocytic lymphohistiocytosis: A case report
Miao SX, Wu ZQ, Xu HG
- 4953** Nasal mucosa pyoderma vegetans associated with ulcerative colitis: A case report
Yu SX, Cheng XK, Li B, Hao JH
- 4958** Amiodarone-induced hepatotoxicity — quantitative measurement of iodine density in the liver using dual-energy computed tomography: Three case reports
Lv HJ, Zhao HW
- 4966** Multisystem involvement Langerhans cell histiocytosis in an adult: A case report
Wang BB, Ye JR, Li YL, Jin Y, Chen ZW, Li JM, Li YP
- 4975** New mutation in EPCAM for congenital tufting enteropathy: A case report
Zhou YQ, Wu GS, Kong YM, Zhang XY, Wang CL
- 4981** Catastrophic vertebral artery and subclavian artery pseudoaneurysms caused by a fishbone: A case report
Huang W, Zhang GQ, Wu JJ, Li B, Han SG, Chao M, Jin K
- 4986** Anastomosing hemangioma arising from the left renal vein: A case report
Zheng LP, Shen WA, Wang CH, Hu CD, Chen XJ, Shen YY, Wang J
- 4993** Bladder perforation caused by long-term catheterization misdiagnosed as digestive tract perforation: A case report
Wu B, Wang J, Chen XJ, Zhou ZC, Zhu MY, Shen YY, Zhong ZX

- 4999** Primary pulmonary plasmacytoma accompanied by overlap syndrome: A case report and review of the literature
Zhou Y, Wang XH, Meng SS, Wang HC, Li YX, Xu R, Lin XH
- 5007** Gastrointestinal stromal tumor metastasis at the site of a totally implantable venous access port insertion: A rare case report
Yin XN, Yin Y, Wang J, Shen CY, Chen X, Zhao Z, Cai ZL, Zhang B
- 5013** Massive gastrointestinal bleeding caused by a Dieulafoy's lesion in a duodenal diverticulum: A case report
He ZW, Zhong L, Xu H, Shi H, Wang YM, Liu XC
- 5019** Plastic bronchitis associated with *Botrytis cinerea* infection in a child: A case report
Liu YR, Ai T
- 5025** Chest, pericardium, abdomen, and thigh penetrating injury by a steel rebar: A case report
Yang XW, Wang WT
- 5030** Monocular posterior scleritis presenting as acute conjunctivitis: A case report
Li YZ, Qin XH, Lu JM, Wang YP
- 5036** Choriocarcinoma with lumbar muscle metastases: A case report
Pang L, Ma XX
- 5042** Primary chondrosarcoma of the liver: A case report
Liu ZY, Jin XM, Yan GH, Jin GY
- 5049** Successful management of a tooth with endodontic-periodontal lesion: A case report
Alshawwa H, Wang JF, Liu M, Sun SF
- 5057** Rare imaging findings of hypersensitivity pneumonitis: A case report
Wang HJ, Chen XJ, Fan LX, Qi QL, Chen QZ
- 5062** Effective administration of cranial drilling therapy in the treatment of fourth degree temporal, facial and upper limb burns at high altitude: A case report
Shen CM, Li Y, Liu Z, Qi YZ

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The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, PubMed, and PubMed Central. The 2020 Edition of Journal Citation Reports® cites the 2019 impact factor (IF) for *WJCC* as 1.013; IF without journal self cites: 0.991; Ranking: 120 among 165 journals in medicine, general and internal; and Quartile category: Q3.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Ji-Hong Liu; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Semimonthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

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

PUBLICATION DATE

October 26, 2020

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INSTRUCTIONS TO AUTHORS

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

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<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>



Primary chondrosarcoma of the liver: A case report

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Supported by National Natural Science Foundation of China, No. 81860304 and No. 81860043; "Thirteenth Five-Year Plan" Science and Technology Project of Education Department of Jilin Province, No. JKH20191141KJ; and Natural Science Research Foundation of Jilin Province for Sciences and Technology, No. 20200201511JC.

Informed consent statement: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict-of-interest statement: All authors have no conflicts of interest.

CARE Checklist (2016) statement: The authors have read the CARE

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Abstract

BACKGROUND

Primary chondrosarcoma of the liver are extremely rare. Moreover, there are few reports focusing on typical clinical symptoms and imaging characteristics. Therefore, the diagnosis of chondrosarcoma of the liver remains a challenge.

CASE SUMMARY

A 59-year-old male was admitted due to a lesion occupying the right liver lobe that was found by physical examination. Magnetic resonance imaging showed a lobular mass with high T2 weighted image and low T1 weighted image with enhanced internal separation and edge in the right liver. He was diagnosed with liver cystadenoma by using magnetic resonance imaging. At 3 mo later, the magnetic resonance scan showed that the mass was enlarged. Laparoscopic liver tumor resection was performed with a pathological diagnosis of liver chondrosarcoma. Then he received a surgical resection for the recurrent lesion. However, intrahepatic and abdominal metastases were found again at 8 mo after the second operation. The patient then received conservative management and is now under follow-up.

CONCLUSION

Primary liver chondrosarcoma generally is presented as lobulated and heterogeneous density/signal, cystic, solid masses without calcification with enhanced edge, internal septa and solid part. The imaging features are closely related to pathology, which may be helpful for clinical diagnosis.

Key Words: Extraskeletal chondrosarcoma; Liver tumor; Differential diagnosis; Case report

Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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Manuscript source: Unsolicited manuscript

Received: May 6, 2020

Peer-review started: May 6, 2020

First decision: May 21, 2020

Revised: June 16, 2020

Accepted: September 2, 2020

Article in press: September 2, 2020

Published online: October 26, 2020

P-Reviewer: Ieni A

S-Editor: Ma YJ

L-Editor: Filipodia

P-Editor: Liu JH



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Core Tip: Primary chondrosarcoma of the liver is extremely rare. Moreover, there are few studies focusing on typical clinical symptoms and imaging characteristics. Therefore, the diagnosis of chondrosarcoma of the liver remains a challenge. This study analyzed the imaging characteristics of a case of primary liver chondrosarcoma to improve the understanding and diagnosis of this disease, which may help to improve the understanding of the disease.

Citation: Liu ZY, Jin XM, Yan GH, Jin GY. Primary chondrosarcoma of the liver: A case report. *World J Clin Cases* 2020; 8(20): 5042-5048

URL: <https://www.wjgnet.com/2307-8960/full/v8/i20/5042.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v8.i20.5042>

INTRODUCTION

Chondrosarcoma is a malignancy characterized by cartilage matrix production by tumor cells, which mainly occurs in cartilage tissue or connective tissue. It more commonly involves the cartilage tissue or connective tissue of long bones of the extremities and pelvis and rarely occurs in soft tissue or other organs, such as the lung, heart, liver, pancreas and kidney, which are known as extraskeletal chondrosarcomas. Extraskeletal chondrosarcomas were first reported by Stout *et al*^[1] and account for 3% of all soft tissue sarcomas. It has been shown that extraskeletal chondrosarcomas have a histopathological characteristic of chondrocyte-like differentiation but no obvious cartilage areas, no connection with bone structure or punctate calcification. Extraskeletal chondrosarcomas are classified as “uncertain differentiated tumors” in the 2013 World Health Organization Soft Tissue Sarcomas^[2]. Extraskeletal chondrosarcomas originating in the liver are rarely observed. Therefore, imaging features of liver chondrosarcoma remain insufficient. In this study, we analyzed the imaging characteristics of a case with primary liver chondrosarcoma to improve the understanding and diagnosis of this disease.

CASE PRESENTATION

Chief complaints

A 59-year-old male was admitted after a physical examination discovered a lesion occupying the right liver lobe.

History of present illness

He had a 40-year history of hepatitis B and received oral “Entecavir” antiviral therapy.

Laboratory examinations

Laboratory tests and tests on tumor markers showed no abnormalities. Ultrasound showed an irregular hypoechoic region in the right posterior lobe of the liver with a poorly smoothed capsule. The uneven echo distribution and a 4.3 cm × 3.8 cm strip-shaped blood flow signal were observed within the hypoechoic region.

Imaging examinations

Magnetic resonance imaging (MRI) showed masses in the posterior lower lobe of the right lobe with a high T2 weighted image (T2WI) signal, low signal rings and separated low signals. The lesion had a low signal in the T1WI positive and negative phases with patches and other signals in the center, had a high diffusion weighted imaging signal with an intralésion patchy low signal and a significantly high signal apparent diffusion coefficient with a low intralésion signal. The lesion had clear boundaries and defoliated edges with a size of 3.1 cm × 4.3 cm × 3.7 cm. No dilatation of the internal bile duct was observed (Figure 1A-C). The edge of the lesion and its internal separating arteries were slightly enhanced during the enhanced scan and

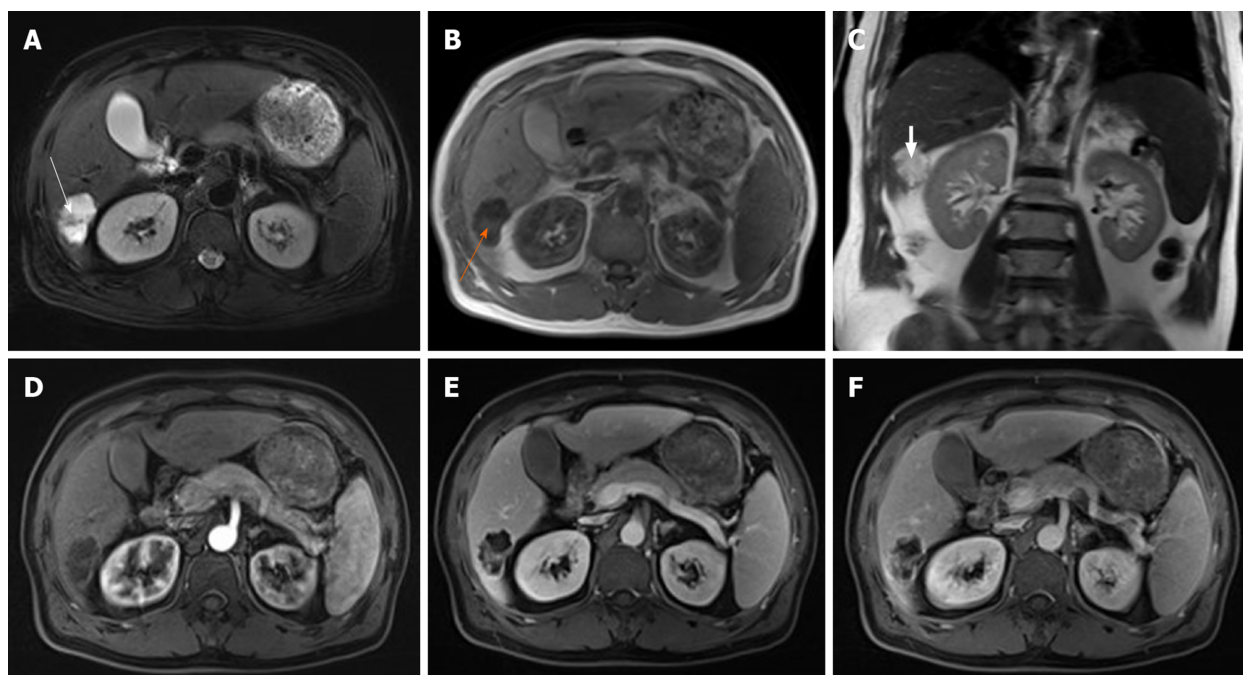


Figure 1 Magnetic resonance imaging at primary clinic visit. A: A magnetic resonance (MR) T2 weighted image (T2WI) axis image showing lobular masses in the posterior lower lobe of the right lobe of the liver with clear borders, a high T2WI signal, intratumor separation with low signal and peripheral capsule with low signal (white arrow); B: An MR T1WI axis image showing the lesion was presented as low signal in T1WI with signals of patches and other (orange arrow); C: A coronal image of T2WI showing that the lesion protruded toward the lower edge of the liver (white arrowhead); D: MR enhanced arterial phase showed that the edge of the lesion and its internal separation are slightly enhanced; E: MR enhanced portal venous phase showed that the edge of the lesion and its internal separation are enhanced; F: MR enhanced delayed stage showed that the edge of the lesion and its internal separation are continually enhanced.

continuously enhanced during portal vein and the delayed phase. No lymphadenopathy was seen in the abdominal cavity (Figure 1D-F).

Diagnostic results of MRI: Occupied lesion in the right lobe of the liver, suggesting cystadenomas. He did not receive further treatment. After 3 mo, he received an MRI scan again, which showed a larger mass with obvious lobulated margin in the right lobe with a size of 4.3 cm × 5.3 cm × 4.9 cm (Figures 2A and B). The relationship between lesions and colonic liver curvature were unclear. Therefore, he was admitted for surgical resection. Intraoperatively, a hard, cauliflower-like mass with size of 5 cm × 6 cm × 5 cm in the lower posterior segment of the right lobe of the moderate sclerosis liver was observed, which adhered to the colonic hepatic mesangium and the right renal capsule.

Pathological gross appearance showed that the cut surface of the tumor was gray-blue, nodular, with a sense of mucus, hard and brittle, and the tumor showed swelling to the surrounding liver tissue. Microscopic pathological examination showed a lobulated tumor with sparsely distributed tumor cells and cartilage pit formation (Figures 3A and B). Tumor cell nuclei were heterogeneous. The cells were mostly mononuclear but double nuclei were also observed. Mitotic cells were commonly observed. There was a transparent cartilage matrix among the cells. The tumor was presented as infiltrating growth to the surrounding liver tissue. The surrounding liver tissue showed nodular cirrhosis. Immunohistochemistry showed vimentin (+), S-100 (+) and Ki67 (60% +) (Figures 3C-E). The pathological diagnosis was chondrosarcoma of the liver.

Emission computed tomography (CT) was performed after surgery and found no space-occupying lesions. Therefore, he was diagnosed as primary liver chondrosarcoma. He had an uneventful postoperative course and received no other treatments. A relapse was found at 8 mo following surgery. He received laparoscopic tumor resection. At 8 mo following the second surgery, he was admitted due to melanosis, fatigue and intermittent midupper abdominal pain.

Physical examination

Physical examination showed a 10 cm × 15 cm mass in the right abdomen. CT scan showed multiple metastases within the liver and abdomen, presenting as multiple cystic masses with marginal lobes and enhanced posterior marginal enhancement, which were like the primary imaging findings (Figures 4A and B). Then, the patient

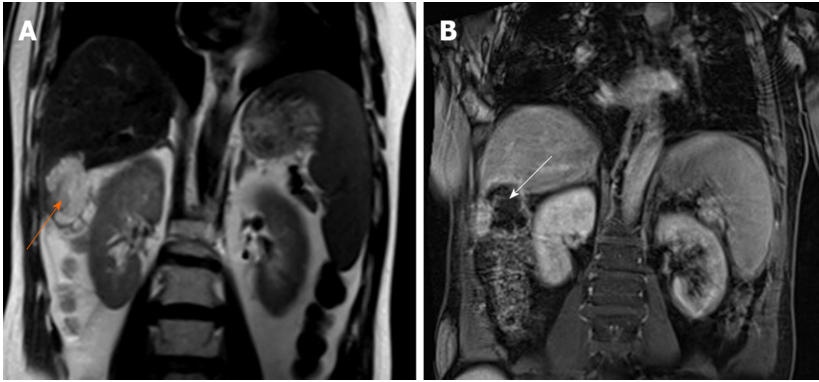


Figure 2 Magnetic resonance imaging after 3 mo. A: A magnetic resonance T2 weighted image coronal image showed that the lesion was larger than before, and the border between the lesion and colonic liver curvature was unclear (orange arrow); B: A magnetic resonance T1 weight image coronal image (white arrow).

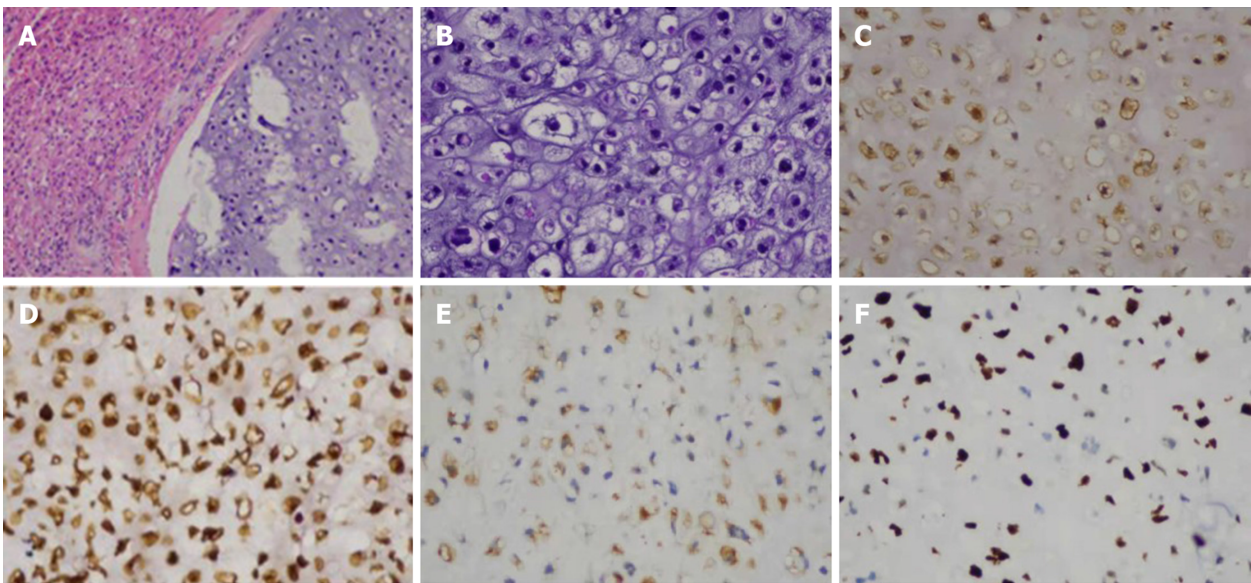


Figure 3 Histopathological evaluation. A: Hematoxylin and eosin staining showed that tumor cells were unevenly distributed with cartilage pits, and the tumor was infiltrating and growing into surrounding liver tissue (magnifications 100 ×); B: Hematoxylin and eosin staining showed tumor cells with abnormal nuclei that were mostly mononuclear though occasionally binuclear (magnifications 200 ×); C: Tumor CK protein positive; D: Tumor vimentin protein positive; E: Tumor S-100 protein positive; F: Tumor Ki-67 immunostaining with 60% Ki-67 positive cells. (C-F immunohistochemistry, magnification 200 ×).

received conservative management and is being followed up until now.

FINAL DIAGNOSIS

Primary liver chondrosarcoma after surgery, multiple metastases within the liver and abdomen.

TREATMENT

Abdominal drainage and anti-inflammatory and liver-protecting symptomatic treatment

OUTCOME AND FOLLOW-UP

The patient was discharged after treatment in good and stable condition, and the

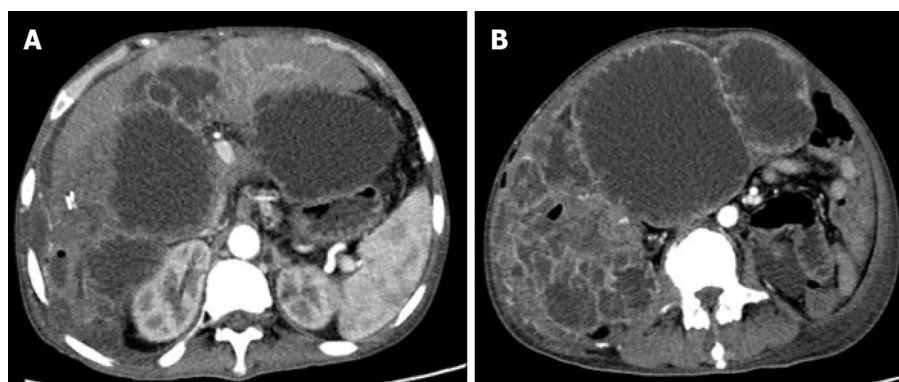


Figure 4 Magnetic resonance imaging at month 8 after the second surgery. A: Computed tomography enhanced arterial phase image showing intrahepatic and abdominal metastases; B: Computed tomography enhanced arterial phase image showing metastatic lesions presenting as multiple cystic masses with enhanced edges.

general condition of the patient was fair after follow-up.

DISCUSSION

Extraskeletal chondrosarcoma is a rare ectopic tumor. Over 90% of patients have a characteristic translocation that involves the *NR4A3* gene on chromosome 9^[3]. Extraskeletal chondrosarcoma, which can be divided into myxoid, mesenchymal and differentiated types^[4], is commonly observed in the deep soft tissues of the proximal limbs with the highest incidence at the age of 50-60 years and more common in males^[5,6].

The incidence of chondrosarcoma in the liver is extremely low. There is no relevant literature abroad. Only two case reports focusing on primary hepatic chondrosarcoma in China were found^[7,8]. Both cases were middle-aged/elderly and male. The main clinical symptom was upper abdominal discomfort but without obvious pain, and both lesions occurred in the right lobe of the liver. Their imaging manifestations were very similar to those of liver cystic tumors, and both were misdiagnosed as hepatic cystadenoma or cystadenocarcinoma before surgery. In this study, the MRI manifestations of liver chondrosarcoma were lobular masses with clear boundaries, obvious high T2WI and diffusion weighted imaging signals with uneven intratumor low signal separation. The lesion had a low T1WI signal without bleeding. There was a capsule around the lesion with a low T2WI signal. After enhancement, the edge of the lesion and its internal separation continually enhanced. CT showed a liquid density mass with lobulated margin and no calcification and same enhancement as that of the MRI.

Chondrosarcoma of the liver should be differentiated from hepatic cystadenoma or cystadenocarcinoma, hepatic hydatid cyst, secondary infection of liver cyst, liver abscess and cystic liver metastases. Hepatic cystadenoma or cystadenocarcinoma is a rare liver epithelial tumor that occurs in middle-aged and elderly females without history of hepatitis and cirrhosis^[9]. The typical CT image characteristics of hepatic cystadenoma or cystadenocarcinoma are single or multiple circular cystic masses with smooth capsule wall, intracapsule separation and calcification^[10]. In the enhanced phase, the posterior capsule wall, the capsule nodules and intracapsule separation are enhanced lightly and moderately but not the capsule fluid, which is very similar to the imaging of the case in our study.

Most patients with hepatic hydatid cyst have a history of nomadic life. The hepatic hydatid cyst is usually presented as single or multiple cystic lesions of the liver with a thick cyst wall and intracyst separation, cyst wall calcification, floating ascus and scolices in the cyst^[11]. For cases with secondary infection of liver cysts, the cyst wall is thickened uniformly and without wall nodules, small air bubbles can appear in the cyst, and a circular edema band is seen around the cyst wall^[12]. The cyst wall is evenly enhanced following the enhancement phase. Clinical manifestations of patients with liver abscess are fever, elevated white blood cell levels and pain in the liver area^[13]. CT of a liver abscess is manifested as circular or quasicircular low-density shadows in the liver parenchyma with even or uneven density^[14]. CT values are higher than water but lower than liver parenchyma. Enhanced scanning shows lesions with a “double ring

sign” and a “petal sign,” which is decreased in the delayed scan phase^[15].

Primary lesions of cystic liver metastases are found in leiomyosarcoma of the digestive tract and colorectal cancer. Most lesions present as multiple lesions with a round shape, while a few are presented as lobulated shape^[16]. When cystic lesions or cystic and parenchymal lesions coexist, the cyst wall can be thin and thick, wall nodules are sometimes seen, and the lesion may have incomplete separation and fluid-fluid level^[17]. The capsule wall, wall nodules and separation can be enhanced during the enhanced phase.

Primary liver chondrosarcoma is extremely rare in the clinic, which also has no specified clinical manifestations. The diagnosis of liver chondrosarcoma mainly depends on pathological evaluation. Imaging characteristics can facilitate the early diagnosis of liver chondrosarcoma. The local recurrence rate of extraskelatal chondrosarcomas is 37%-48%^[5], with a metastasis rate of 50%^[5]. Extraskelatal chondrosarcoma is not sensitive to chemotherapy and radiotherapy. Currently, early surgical resection is considered the most effective treatment method for extraskelatal chondrosarcomas^[18,19]. In this study, the patient was misdiagnosed as a hepatic cystadenoma due to lack of understanding of the imaging manifestations of liver chondrosarcoma. The lesion was increasingly enlarged, and surgical resection was performed after 3 mo. Thereafter, he received a surgical resection for recurrent lesion. However, intrahepatic and abdominal metastases were found again after the second operation. Thereafter, conservative treatments and regular follow-up was administrated.

CONCLUSION

In conclusion, primary liver chondrosarcoma is very rare, and it is difficult to accurately diagnose only by imaging findings. The diagnosis is confirmed mainly based on pathological evaluation and immunohistochemical phenotypes. The imaging characteristics of the patient with primary liver chondrosarcoma in this study may help to improve the understanding of the disease.

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