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Liver Solitary Fibrous Tumour with Local Recurrence and Metastasis: A Case Report

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Abstract

Solitary fibrous tumour (SFT) of the liver is an extremely rare tumour, and only four cases of liver SFT with local recurrence and metastasis have been reported in the English scientific literature. In this report, we discuss a case of a 66-year-old woman who was misdiagnosed as hepatic cellular carcinoma (HCC). The patient successfully underwent a resection of the tumour, and the diagnosis was confirmed as SFT on histopathology. However, the patient developed a local recurrence and pulmonary metastasis. Our case shows that primary malignant SFT may occur in the liver and long-term follow-up is advised.

Keywords: Solitary fibrous tumour; Hepatic neoplasm; Malignancy; Metastasis; Case

Introduction

The solitary fibrous tumour (SFT) is a rare spindle-cell neoplasms arising from mesenchymal soft tissue, which occurs most frequently in pleura [1], occasionally the peritoneum [2] and other anatomic location. However, the liver SFT is exceedingly rare and only 4 cases of liver SFT with local recurrence and metastasis have been reported in English scientific literature [3]. Liver SFT may be easily misdiagnosed because it has nonspecific laboratory examination and imaging performance. Herein, we report a malignant tumour of the liver histologically diagnosed as SFT with local recurrence and pulmonary metastasis.

Case Presentation

A 66-year-old woman presented with upper abdominal pain, anorexia, and weight loss for one month, was referred to our hospital on August 13, 2019. The patient described no nausea, no vomiting, no fever, nor jaundice or evidence of infectious history such as hepatitis and tuberculosis. On physical examination, the abdomen was soft; a slight-painful liver edge descended to 9 cm below the right costal margin. There were no abnormalities in laboratory examinations. Serum tumour markers (CA199, AFP, and CEA) were all negative. Computed tomography (CT) scan revealed a large mass in the right lobe of liver measuring 12.9×14.3×13.2 cm (Figure 1). On magnetic resonance (MR) scan, the mass was predominantly low signal at T1WI (high signal represents haemorrhage) and inhomogeneous hyper intensity on T2WI (the liquid-liquid level was revealed). Diffusion weighted imaging (DWI) showed diffusion restriction. Contrast-enhanced T1-weighted MR imaging of gadopentetic acid (GD-DTPA) showed heterogeneous enhancement in the arterial phase and persistent enhancement in the portal and late phases (Figure 2). The resection specimen was unclear and measured 22 × 18 × 13cm in size with the liver metastasis in the caudate lobe. Postoperative pathology confirmed a malignant solitary fibrous tumour of the liver (Figure 3). Histological examination: Ki67 (40 % +), Vimentin (+), Desmin (+), CD34 (-), CD99 (+), Hep (-), S-100 (-), SOX10 (-), STAT-6 (+). The patient recovered well and was discharged on 5 September 2019. Thereafter, a CT examination was performed every 6 months and there was no evidence of recurrence. However, on May 21, 2021, tumour recurrence and a new pulmonary metastasis (left lung metastasis)

were found at re-examination (Figure 4). The patient was discharged after symptomatic treatment and followed up was done for one year with no further symptoms reported.

Discussion

Solitary fibrous tumour (SFT) is a rare neoplasm originating from mesenchymal cells, first reported by Klemperer and Rabin in 1993 [4,5]. SFTs have been described in different tissues and organs, most often in the pleura, as well as in the peritoneum, respiratory tract, mediastinum, lung and others [6,7].

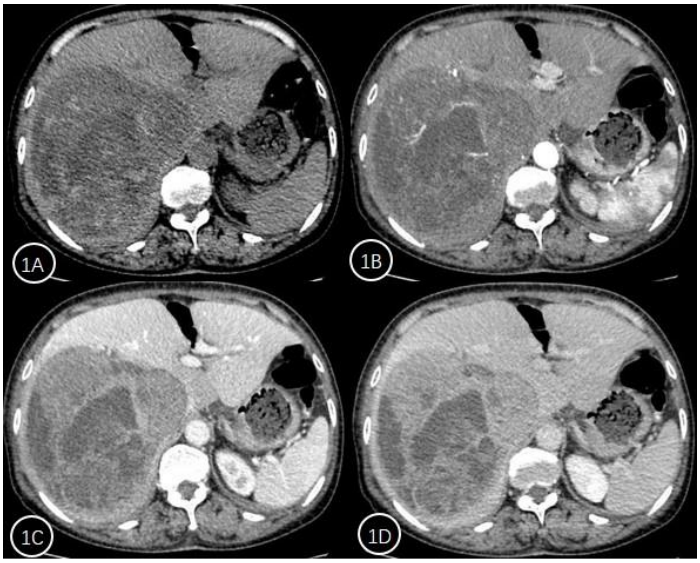

Figure 1: *Abdominal computed tomography scan of the tumor. (A) Non-contrast computed tomography image showing a large mass with an unclear border and irregular morphology in the right lobe of the liver. (B) Moderate inhomogeneous enhancement was found in the arterial phase, with an obviously enhanced staghorn shape vessel. (C) Further enhancement was found in the portal phase. The right branch of the portal vein was compressed, and the boundary was unclear. (D) During the late phase, the lesion had iso-enhancement comportment. No enhancement was observed in the necrotic area.*

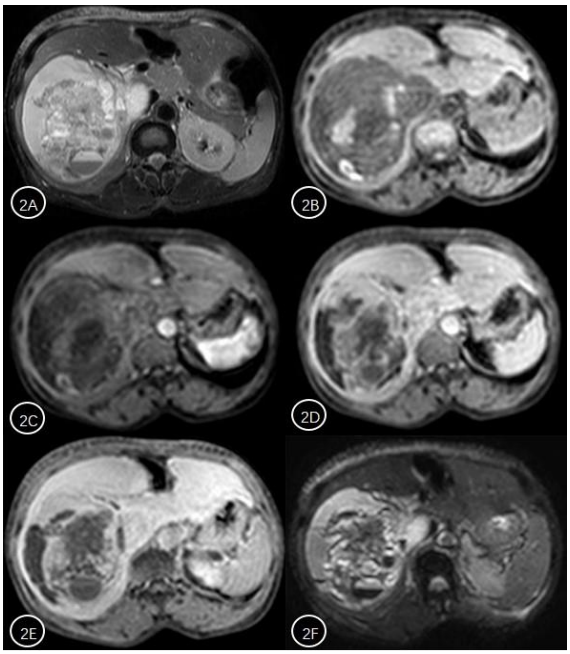

Figure 2: *MRI findings of malignant solitary fibrous tumor of the liver. (A) T2WI showed a heterogeneous hyperintense lesion with an unclear margin and irregular morphology in the right lobe of the liver. Liquid-liquid level and hemosiderin deposition were seen in the lesion. (B) T1WI showed hypointensity, internal hemorrhage showed patchy high intensity. (C-E) GD-DTPA enhanced scan showed inconsistent enhancement in the arterial phase and persistent enhancement in the portal and late phases. No enhancement was observed in the necrotic area. (F) DWI demonstrated restricted diffusion.*

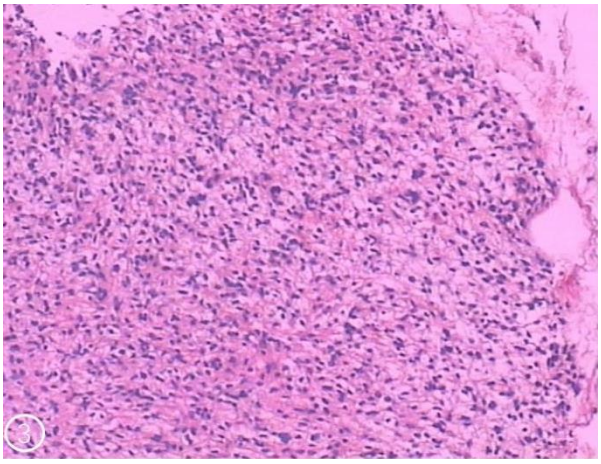

Figure 3: *Histological examination of malignant solitary fibrous tumor of the liver. The tumor was composed of short spindle-shaped cells and the nuclei were round or oval. The cell showed mild nuclear atypia and nuclear mitotic.*

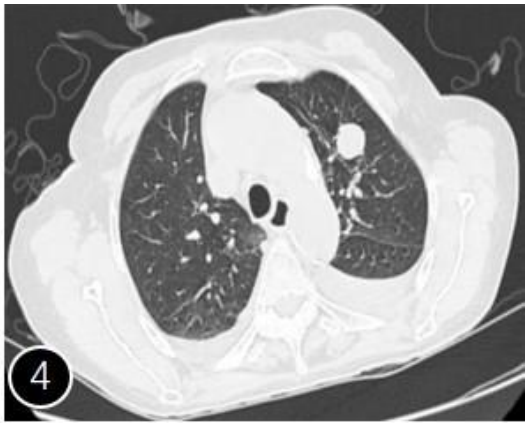

Figure 4: *Chest CT images of patient with malignant solitary fibrous tumor of the liver was reviewed. A 1.7 × 1.8 cm mass was seen in the upper lobe of the left lung with a clear boundary, accompanied by pleural effusion on both sides.*

There are few systematic and comprehensive reports on SFT of the liver. Although the majority are benign, approximately 10-15% of SFTs exhibit malignant behaviours, including recurrence and metastasis [8,9]. In the 2019 WHO classification of tumours of the digestive system, SFT was a mesenchymal tumour with malignant potential [10]. The mean age of diagnosis for liver SFT was 57.1 years, and females predominated (1.4:1) [11]. The clinical presentation is closely associated with tumour size, occurrence place, benign and malignant. About 80 % of the patients were asymptomatic when they came to the hospital and were occasionally found during regular physical examination [12]. With tumour enlargement or compression of adjacent organs, patients will be accompanied by nausea, vomiting, and right upper abdominal dull pain, shortness of breath, weight loss and fatigue. According to previous literature, hypoglycaemia also occurs in a few patients because the tumour produces and secretes insulin-like growth factors (IGF-2) [13]. There was no significant difference in clinical symptoms between patients with malignant liver SFT and patients with benign liver SFT. No abnormalities were found in laboratory examination and serum tumour markers, and so was this case. At present, there is no specific imaging diagnostic method for SFT of the liver. Although ultrasound and radiological assessments are usually not characteristic, they are still the main method of preoperative diagnosis of SFT. Ultrasound showed an unevenly solid mass with no uniform internal echo demonstrating alternate distribution of hypo- and hyper cellular areas [14]. The unenhanced CT scan showed soft tissue density on solid part of SFT; Contrast-enhanced CT scan demonstrated inhomogeneous continuous enhancement, cystic and necrotic area were not enhanced. The branch vessels in the tumour were obviously staghorn-shaped enhancement. The MRI findings were similar to CT description. On MR imaging, liver SFT had low signal on T1WI and intermediate or slightly high signal on T2WI. After GD-DTPA injection, compared with the normal liver parenchyma, SFT was mildly enhanced in the solid part of tumour on arterial phase and delayed wash-out on late phase which reflects abundant collagen fibers [15]. Diffusion-weighted imaging show restriction with hyper intense signal. SFT

of liver is easily misdiagnosed. Our patient was diagnosed as liver malignant tumour, including hepatocellular carcinoma (HCC) and intrahepatic cholangiocellular carcinoma (ICC). Liu et al. reported that collagen fibres showed low signal on T2WI, which may be great of value in the diagnosis of SFT; multiphase enhanced scanning helps to improve the diagnostic accuracy of liver SFT [16]. Diagnosis of SFT mainly depends on histopathological and immunohistochemically results. Microscopically, SFT had a pattern less architecture containing hypo- and hyper cellular areas of short spindle-cells, separated by collagenous stroma with staghorn-shaped vessels (hemangiopericytoma-like structure) [5,15,17]. Mitosis is occasionally detected, but nuclear pleomorphism or atypia is rare [18]. Immunohistochemistry showed positive expression of CD34, CD99, vimentin and Bcl-2 in SFT of liver [6]. CD34 is a biomarker of mesenchymal tumour cells and 5-10 % of SFT has no response to CD34, lacking specificity and sensitivity [19]. Malignant SFT can show deficiency of CD34 expression due to overexpression of p53 and p16 genes [20]. For benign SFT, Ki-67 was positive and less than 5 %; when Ki-67 is higher than 5 %, it suggests malignant potential [21,22]. According to the latest research, STAT-6 has been considered a highly sensitive and specific immunohistochemically marker that could help diagnose SFT [23]. In this patient, CD34 was negative but STAT-6 appeared positive, which enabled us to distinguish SFT from other tumours. Up to date, no scientific and effective management guidelines had been established, and surgical resection is the main treatment of liver SFT patients. After surgical resection, the survival rate was over 90 % without recurrence [24]. All reported patients with liver SFT accepted surgery treatment, including malignant cases. Previous reports have shown that patients have no benefit from radiotherapy or chemotherapy [21,25-28]. The biological behaviour of SFT is unpredictable, so the prognosis exist uncertainty. Therefore, the most important factor affecting the prognosis of patients with liver SFT is complete resection of the tumour, and the tumour margin is free of involvement [29]. England et al reported the criteria about malignant SFT: large tumour size (> 10 cm), tumour necrosis and haemorrhage, cellular atypia, mitotic changes (>4/10 HPFs), nuclear pleomorphism, metastasis. After surgical resection, 26.7 % of patients with malignant SFT had local recurrence within 9 months to 6 years, and 53 % had distant metastasis within 1 month to 6 years [3]. This case also had recurrence and pulmonary metastasis 1 year after surgery. Malignant SFT possesses invasive potential, and systematic long-term follow-up of patients is necessary. Liver SFT is a rare neoplasm. Nonspecific laboratory findings and atypical radiological manifestations lead to a high misdiagnosis rate. Complete surgical excision is the treatment of choice for liver SFT. The diagnosis is mainly confirmed by histological and immunohistochemically analysis of the resected tumour. Considering the late recurrence or metastasis, a long-term follow-up of these patients is advised in all liver SFT patients.

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