Ex situ hepatectomy and liver autotransplantation for a treating giant solitary fibrous tumor: A case report

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Abstract. A solitary fibrous tumor (SFT) is a rare mesenchymal tumor. *Ex situ* hepatectomy and liver autotransplantation are novel methods for the treatment of complicated liver tumors, for example, those involving vascular structures, including the inferior vena cava, which are unresectable by conventional approaches. The present study describes a rare case of a massive hepatic SFT in a 32-year-old female who underwent *ex situ* hepatectomy and liver autotransplantation to achieve a radical resection. The surgery was without complications. Post-operative histopathological and immunohistochemical examinations revealed an SFT of the liver. The patient was discharged 29 days after the surgery with fully recovered liver function. The routine check-up 3 months after surgery indicated normal liver function and no evidence of recurrence. Additionally, an exhaustive review of available literature was performed to provide a complete overview of the current status of SFTs. In summary, the present study found that *ex situ* hepatectomy and liver autotransplantation are suitable surgical techniques for treating a giant SFT, as well as other liver neoplasms that are considered unresectable by conventional surgery.

Introduction

A solitary fibrous tumor (SFT) is a rare mesenchymal tumor, which can be found in different locations within the human body (1). SFTs were first reported in the pleura and subsequently in the peritoneum, thymus, orbit, pericardium, meninges, spinal cord, and the parotid and thyroid glands (2). A tumor of this type in the liver is an extremely rare occurrence. The majority of SFTs are considered benign, while <20% are reported to be malignant and are accompanied by tumor invasion and metastasis (3). Surgical resection is the main approach for SFT treatment. Nevertheless, in certain cases where vascular structures are involved, conventional surgery may not be practical.

Ex situ hepatectomy and liver autotransplantation are novel methods for treating complicated liver tumors that are unresectable by conventional approaches, including liver transplantation, vascular reconstruction, organ perfusion, extended hepatic resection and hemodynamic management, which are considered to be some of the most complicated, difficult and risky types of surgeries. In this maneuver, the whole liver is removed and perfused with cold preservation solution. The tumor is resected *ex situ* on the operating table and the remaining liver is orthotopically implanted. The *ex situ* liver resection was first described by Pichlmayr *et al* (4) as a novel surgical procedure to treat a bilateral liver leiomyosarcoma. To date, only a few cases of *ex situ* hepatectomy and liver autotransplantation have been described worldwide (5,6). Furthermore, to the best of our knowledge, there are no reports on adopting *ex situ* hepatectomy and liver autotransplantation as an approach for treating hepatic SFTs. The present case study reports the first case of a giant SFT involving the inferior vena cava (IVC) of the liver in a 32-year-old female who was treated with *ex situ* hepatectomy and liver autotransplantation.

Case report

A 32-year-old female suffering from repeated abdominal distension for 3 years was found to have an oversized mass in the right hepatic lobe using a B-scan ultrasound, and was admitted on July 7, 2017 to the Outpatient Department of the First Affiliated Hospital, School of Medicine, Zhejiang University (Hangzhou, China). A physical examination revealed a large mass in the right hypochondrium, causing local discomfort. No aberration was observed regarding the medical history

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of the patient. In addition, no elevation of tumor markers, including α-fetoprotein (AFP; ARCHITECT AFP Reagent kit; cat. no. 3P36-30; Abbott Pharmaceutical Co., Ltd., Lake Bluff, IL, USA), carcinoembryonic antigen (CEA; ARCHITECT CEA Reagent kit; cat. no. 7K68-32; Abbott Pharmaceutical Co., Ltd.), cancer antigens (CA) 125 (ARCHITECT CA 125 II Reagent kit; cat. no. 2K45-35; Abbott Pharmaceutical Co., Ltd.) and 19-9 (ARCHITECT CA 19-9 XR Reagent kit; cat. no. 2K91-38; Abbott Pharmaceutical Co., Ltd.), was observed, as detected using immunofluorescence assays, according to the manufacturer's protocols. Hepatitis virus markers were all negative, as detected using an immunofluorescence assay (ARCHITECT Reagent kits; cat. nos. 6C36-44, 8L44-30, 6C34-35, 6C32-20 and 7C18-34; Abbott Pharmaceutical Co., Ltd.), according to the manufacturer's protocol. The patient underwent an enhanced abdominal computed tomography (CT) scan, which revealed a giant, hypodense, heterogeneous, cystic-solid lesion (20.0x16.0 cm), with irregular contrast enhancement, causing compression of neighboring structures. Dilation of the intrahepatic biliary ducts was also noted (Fig. 1). A CT arteriography scan showed that the mass was supplied with blood by the left and right hepatic arteries. The right, middle and left hepatic veins were compressed by the mass and there was indication of a filling defect in the IVC. A magnetic resonance imaging scan was also performed, revealing an oversized, heterogeneous liver mass (21.2x19.7 cm). A heterogeneous high signal in the T2-weighted image and a slightly low signal in the T1-weighted image were observed. All results indicated that the lesion was malignant and a mesenchymal tumor was suspected.

In view of the young age of the patient, an ex situ hepatectomy and liver autotransplantation were performed on October 17, 2017. During the surgery, the second hepatic portal was difficult to expose and the sternum was therefore removed with the assistance of a cardiothoracic expert. An intra-operative frozen section examination indicated the proliferation of fibrotic tissue, and the tumor was considered to be benign. Consequently, the tumor was resected on the bench. A 26-mm artificial blood vessel was utilized to establish an end-to-side portocaval shunt during the anhepatic period and the liver remnant was autotransplanted in a piggyback fashion. The surgery was without complications and lasted for 18 h, with an anhepatic period of 5 h.

The resected specimen was well-delimited and measured 19.0x19.5x14.0 cm in size and 3.964 kg in weight. The tumor presented with a grayish-white interlaced appearance in the section plane, and was comprised of spindle-shaped cells infiltrated with lymphocytes, plasmocytes and mastocytes (Fig. 2). The immunohistochemical (IHC) examination was performed on paraffin-embedded specimens fixed in 4% buffered neutral formalin at 37°C for 24 h. Paraffin-embedded sections (3 μm) were deparaffinized and and endogenous peroxidase was inactivated with 15% H_2O_2-Methanol for 5 min at room temperature. The sections were then incubated with primary antibodies at 37°C for 30 min, according to the manufacturers' protocols. Subsequently, the sections were incubated with Dako REALREEnVision Detection system, Peroxidase/DAB, Rabbit/Mouse (cat. no. K5007; Dako; Agilent Technologies, Inc., Santa Clara, CA, USA) for 30 min at room temperature. Results revealed that the sections were positive for cluster of differentiation 117 (CD117; cat. no. 790-2951; dilution

1:100; Roche Diagnostics, Basel, Switzerland), CD34 (cat. no. M-0117; dilution 1:50; Shanghai Changdao Biotechnology Co., Ltd., Shanghai, China) and Caldesmon (cat. no. ZA-0535; dilution 1:100; OriGene Technologies, Inc., Beijing, China). In addition focal expression of smooth muscle actin (cat. no. ZM-0003; dilution 1:100; OriGene Technologies, Inc.) and progestrone receptor (cat. no. NCL-L-PGR-312; dilution 1:300; Leica Biosystems, Ltd., Milton Keynes, UK) expression was detected. However, sections were negative for S-100 protein (cat. no. ZA-0225; dilution 1:200; OriGene Technologies, Inc.), desmin (cat. no. ZM-0091; dilution 1:100; OriGene Technologies, Inc.), deletions of G-rich-1 (cat. no. ZM-0371; dilution 1:200; Zhongshanjinqiao Bio-Reagent Company), cytokeratin (cat. no. M-0349; dilution 1:100; Shanghai Changdao Biotechnology Co., Ltd.), β-catenin (cat. no. ZM-0442; dilution 1:100; OriGene Technologies, Inc.), anaplastic lymphoma kinase (cat. no. ALK-L-CE-H; dilution 1:150; Leica Biosystems, Ltd.), estrogen receptor (cat. no. NCL-L-ER-6F11; dilution 1:100; Leica Biosystems, Ltd.), epithelial membrane antigen (cat. no. ZM-0095; dilution 1:200; Zhongshanjinqiao Bio-Reagent Company), wilms tumor type 1 (cat. no. ZM-0269; dilution 1:100; Zhongshanjinqiao Bio-Reagent Company) and CD10 (cat. no. NCL-L-CD10-270; dilution 1:100; Leica Biosystems, Ltd.). The sections were evaluated under a light microscope at x200 magnification. The mean proliferative index was 5% in the hypercellular areas, as determined by detection of antigen KI-67 (Ki-67; cat. no. ZM-0166; dilution 1:100; Zhongshanjinqiao Bio-Reagent Company). The Ki-67 proliferative index was examined under a light microscope (BX53; Olympus Corporation, Tokyo, Japan), and was evaluated by experienced pathologists. A total of 500 tumor cells were counted both at the center and at the periphery of the tumor. The examination was repeated at least twice and the mean Ki-67 proliferative index was calculated. At least 10 mm of the margins of the resected specimen were tumor-free. Based on the pathological and IHC characteristics, the lesion was identified as an SFT.

On post-operative day 9, the patient underwent pleurocentesis and received drainage tubes due to moderate right pleural effusion, which were successfully removed 16 days later. The patient recovered fully and was discharged on post-operative day 29. At the 1- and 3-month follow-ups, the patient had normal liver function and there was no evidence of recurrence. Future follow-up will be performed every 6 months.

Discussion

SFTs are mesenchymal neoplasms that typically occur in the thoracic or pleural cavities. The SFTs found in the liver may derive from the intra-hepatic connective tissue, for example Glisson's capsule or conjunctive tissue, which are rare types of liver neoplasm (7). A search of the English literature on 'Solitary Fibrous Tumour of the Liver' was conducted on PubMed (https://www.ncbi.nlm.nih.gov/pubmed/) and Google (https://www.google.com/). All published English articles, case reports and literature reviews, and their reference lists, were reviewed. The results indicated that only 88 cases of SFTs of the liver have been reported since 1958 (7-71). The main characteristics of these cases are listed in Table I. The mean age of onset was 57.1 years (range, 16.0-87.0 years)
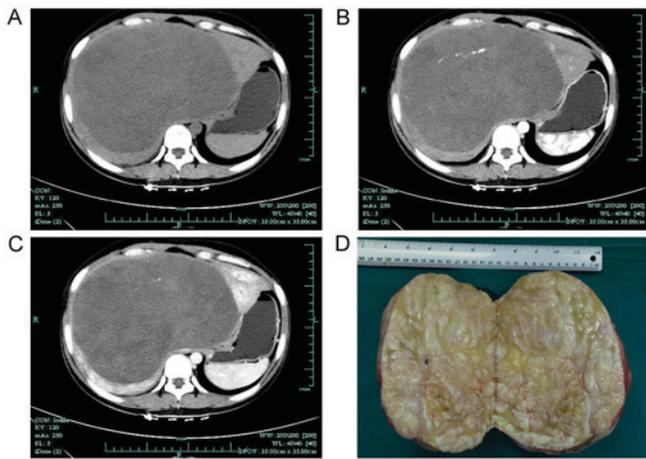
Figure 1. Abdominal computed tomography scan prior to surgery and gross examination of the specimen. Abdominal computed tomography scans [(A) plain, (B) arterial and (C) portal phase] revealed a large, hypodense, heterogeneous, cystic-solid lesion measuring 20.0x16.0 cm, with irregular contrast enhancement, causing compression of the neighboring structures. (D) The resected specimen was well-delimited and measuring ~19x19.5x14 cm, with a grayish-white interlaced appearance of the section plane.
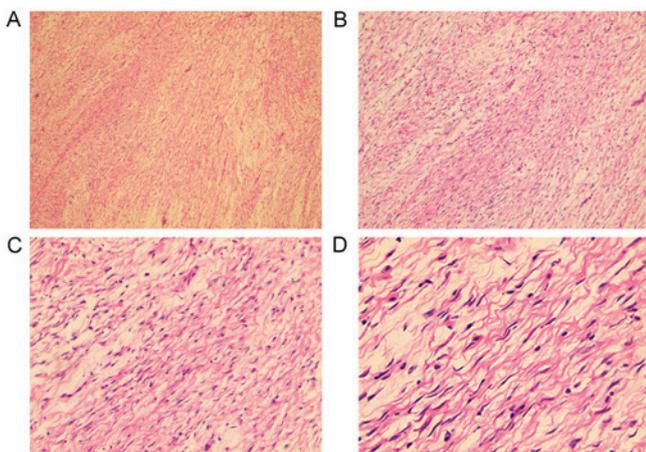

Figure 2. Histological examination showed that the tissues consisted of spindle-shaped cells that were infiltrated with lymphocytes, plasmocytes and mastocytes. The images are of hematoxylin and eosin staining at (A) x50, (B) x100, (C) x200 and (D) x400 magnification.

and the lesion appeared more commonly in female patients (ratio 1.4:1), with a mean tumor diameter of 18.2 cm (range, 1.5-30.0 cm). The median follow-up time for these cases was 13.5 months. The prolonged course and biological features of SFTs resulted in a relatively late age of onset and large lesion size. The patient in the present study was a 32-year-old female with a single lesion measuring ~19.0 cm; these characteristics were consistent with the cases listed in Table I.

Due to the lack of overt or specific symptoms, patients with SFTs are usually diagnosed coincidentally during a check-up or at a late stage when masses are large enough to produce discomfort by invading or compressing adjacent structures. In the present case, the patient had complained of repeated abdominal distension for 3 years due to the compression of neighboring structures by the lesion. With the exception of the presence of non-islet cell tumor hypoglycemia syndrome observed in

13 cases (9,16,17,19,22,25,28,31,36,48,60,69,70), the results of laboratory tests for routine biochemical parameters, including aminotransferases or tumor markers, appear normal or slightly, non-specifically altered (33), as was the case with the present patient. Although imaging tests can provide physical characteristics of lesions for clinical analysis, including location and size, the radiological features of an SFT lack specificity, suggesting that none of the aforementioned tests are conclusive.

The gold standard for diagnosing an SFT remains as histopathological analysis combined with IHC examination. Fine-needle aspiration cytology (FNAC) is commonly performed to acquire lesion tissue for pathological diagnosis prior to surgery. Nevertheless, FNAC may be misleading or inconclusive pertaining to the diagnosis of an SFT (37). Percutaneous liver biopsy may not provide the definitive diagnosis, while increasing the risk of tumor growth and implantation (53). Histopathological findings in the present case were typical for an SFT: Diffusive proliferation of spindle-shaped mesenchymal cells with oval-fusiform nuclei along with fibrocollagenous stroma; no prominent cellular atypia, infiltrative margins or other indicative evidence of malignancy; and tumor cells of all reported cases with an SFT, including the present case, were positive for CD34, which may differentiate an SFT from other spindle cell neoplasms (12). CD99, B-cell lymphoma 2 or vimentin have also shown to be positive in almost half the cases (14-19-21,24-27,29-33,36-48,50,52-71).

Radical surgical resection is the main treatment approach. Among the reported cases listed in Table I, 85.2% (75/88) patients received radical surgical resection and on average had ≥3 segments removed owing to the size of the SFTs. Other treatments, such as chemotherapy, which was adopted recently in two patients listed in Table I, resulted in only 4-5 months survival following treatment. Therefore, Makino *et al* (67) suggested that due to the unclear effects of other treatments, including chemotherapy and radiotherapy, hepatic resection with clear margins is highly recommended in patients with large SFTs, considering the risk of urgent symptoms and the potential for malignancy. Compared with the previously reported cases, the tumor reported in the present study was difficult to resect using the conventional approach due to the advanced extent of the lesion therefore, novel techniques may be required.

Even with the vast improvement of surgical techniques, hepatic lesions abutting hepatic veins or the confluence of the IVC, as well as large centrally located lesions, fail to be managed by conventional surgery. In such cases, liver transplantation may be indicated (26). However, no liver transplantations have been performed to date for treating SFTs. In the present case, the second hepatic portal was compressed by the neoplasm, causing obstruction of the outflow tract and liver congestion. Full exposure of the second hepatic portal was achieved only after the sternum was removed. The vascular reconstruction and extended hepatic resection were extremely difficult to achieve by *in situ* surgical resection. Given the young age of the patient, the alternative of *ex situ* hepatectomy and liver autotransplantation was implemented. The surgery was performed successfully and the patient recovered without further complications. To the best of our knowledge, this is the first case report describing the adoption of *ex situ* hepatectomy and liver autotransplantation to treat a liver SFT, otherwise unresectable by standard surgery.

Table I. Main characteristics of reported liver solitary fibrous tumor cases.

First author, year	Age, years	Sex	Lobe	Size, cm	Hypo	Treatment	IHC	Follow-up	(Refs.)
Edmondson, 1958	16	F	R	23x17	N	Resection	N/A	24 months	(8)
	N/A	N/A	R	5x5	N	Resection	N/A	N/A	
Nevius and Friedman, 1959	56	M	R	15x15	Y	Radiation	N/A	Succumbed after 2 days	(9)
Ishak, 1976	62	M	L	24	N	Resection	N/A	N/A	(10)
	62	F	L	23x20x13	N	Resection	N/A	Succumbed during surgery	
Kim and Damjanov, 1983	27	F	L	27x23x15	N	Resection	N/A	6 months	(11)
Kottke-Marchant *et al*, 1989	84	F	L	15x9x8	N	Resection	V$^+$	29 months	(12)
Kasano *et al*, 1991	39	F	L	18x10x18	N	Resection	N/A	53 months	(13)
Barnoud *et al*, 1996	50	M	R	17x15x11	N	Resection	CD34$^+$, V$^+$	N/A	(14)
Levine and Rose, 1997	57	M	L	10x18x8	N	Resection	CD34$^+$, V$^+$	38 months	(15)
Guglielmi *et al*, 1998	61	F	R	20x16x10	Y	Resection	CD34$^+$, V$^+$	72 months	(16)
Lecesne *et al*, 1998	69	F	L	N/A	N	Resection	CD34$^+$, V$^+$	12 months	(17)
Bejarano *et al*, 1998	49	M	L	17x12x10	N	Resection	CD34$^+$, V$^+$	15 months	(18)
Moran *et al* 1998	62	F	N/A	23x20x13	N	Resection	CD34$^+$, V$^+$	N/A	(19)
	34	F	N/A	2x0.5	N	Nil	N/A	Incidental (autopsy)	
	57	F	N/A	24x19x11	N	Resection	CD34$^+$, V$^+$	N/A	
	32	M	N/A	12x9x7	N	Resection	CD34$^+$, V$^+$	N/A	
	68	F	N/A	17x17	N	Resection	CD34$^+$, V$^+$	Succumbed after 2 days	
	83	F	R	18	Y	Resection	CD34$^+$, V$^+$	Succumbed after 6 days	
	72	F	L	9	N	Resection	CD34$^+$, V$^+$	12 months	
	62	M	L	24	N	Resection	CD34$^+$, V$^+$	N/A	
	50	F	N/A	3x2x1.5	N	Resection	CD34$^+$, V$^+$	N/A	
Fuksbrumer *et al*, 2000	40	F	R	14-17	N	Resection	CD34$^+$, V$^+$, Bcl-2$^+$	N/A	(20)
	71	F	R	14-17	N	Resection	CD34$^+$, V$^+$, Bcl-2$^+$	N/A	
	80	M	R	14-17	N	Nil	CD34$^+$, V$^+$, Bcl-2$^+$	N/A	
Yilmaz *et al*, 2000	25	F	R	32x30	N	Resection	V$^+$	6 months	(21)
Lin *et al*, 2001	75	M	R	21x20x18	Y	Resection	CD34$^+$	11 months	(22)
Gold *et al*, 2002	N/A	N/A	N/A	N/A	N	N/A	N/A	N/A	(23)
	N/A	N/A	N/A	N/A	N	N/A	N/A	N/A	
Neeff *et al*, 2004	63	F	R	30x12x19	N	Resection	CD34$^+$, V$^+$	6 months	(24)
Chithriki *et al*, 2004	76	F	R	20x15x16	Y	Resection	CD34$^+$, Bcl-2$^+$	11 months	(25)
Vennarecci *et al*, 2005	65	M	R	30x28x14	N	Resection	CD34$^+$, V$^+$	30 months	(26)
Moser *et al*, 2005	73	F	R	35x20x15	Y	Resection	CD34$^+$, V$^+$, Bcl-2$^+$	N/A	(27)
Ji *et al*, 2006	42	F	R	6x5x5	Y	Resection	CD34$^+$	N/A	(28)
Lehmann *et al*, 2006	63	F	R	N/A	N	Resection	CD34$^+$	96 months	(7)
Nath *et al*, 2006	61	F	R	21x14.5x30	N	Resection	CD34$^+$, V$^+$	10 months	(29)
Terkivatan *et al*, 2006	74	M	L	24x21x15	N	Resection	CD34$^+$, CD99$^+$, V$^+$, Bcl-2$^+$	12 months	(30)
Chan *et al*, 2007	70	M	R	27x24x12	Y	Resection	CD34$^+$, CD99$^+$, V$^+$, Bcl-2$^+$	9 months	(31)

Table I. Continued.

First author, year	Age, years	Sex	Lobe	Size, cm	Hypo	Treatment	IHC	Follow-up	(Refs.)
Obuz et al, 2007	52	M	L	10x11x12	N	Resection	CD34+, V+	22 months	(32)
Perini et al, 2008	40	F	L	N/A	N	Resection	CD34+, V+	49 months	(33)
Weitz et al, 2007	N/A	N/A	N/A	N/A	N	Resection	N/A	N/A	(34)
	N/A	N/A	N/A	N/A	N	Nil	N/A	N/A	
	N/A	N/A	N/A	N/A	N	Nil	N/A	N/A	
Kandpal et al, 2008	45	F	R	N/A	N	Resection	CD34+	N/A	(35)
Fama et al, 2008	68	M	R	N/A	Y	Resection	CD34+, V+	25 months	(36)
Korkolis et al, 2008	82	F	L	18x15x8	N	Resection	CD34+, V+, Bcl-2+, desmin+	21 months	(37)
Chen et al, 2008	71	M	R	8.7x5.5x8.5	N	Resection	CD34+, CD99+, Bcl-2+	9 months	(38)
El-Khouli et al, 2008	68	F	L, R	15x10.5x13	N	TACE	CD34+, V+	N/A	(39)
Hoshino et al, 2009	30	F	R	6.7x4.5x4	N	Nil	CD34+, Bcl-2+	6 months	(40)
Novais et al, 2010	34	F	R	25x23x13	N	Resection	CD34+, V+	24 months	(41)
Brochard et al, 2010	54	M	R	17	N	Resection	CD34+, V+, desmin+, actin+	72 months	(42)
Haddad et al, 2010	62	M	L	N/A	N	Resection	CD34+	N/A	(43)
	45	F	R	7.4x5.9x5.4	N	Resection	CD34+, V+, Bcl-2+	N/A	
Park et al, 2011	51	F	L	N/A	N	Resection	N/A	N/A	(44)
Peng et al, 2011	24	F	R	30x17x15	N	Resection	CD34+, V+, Bcl-2+	Succumbed after 16 months	(45)
Sun et al, 2011	59	M	L	9x7x6	N	Resection	CD34+, CD99+, V+, Bcl-2+	24 months	(46)
Patra et al, 2012	34	F	L	14.5x10x8	N	Resection	CD34+, V+, Bcl-2+	48 months	(47)
Radunz et al, 2012	85	F	L	N/A	Y	Resection	CD34+, Bcl-2+	N/A	(48)
Belga et al, 2012	66	F	R	N/A	N	Resection	CD34+	30 months	(49)
Morris et al, 2012	23	F	R	27x 23.5x4	N	Resection	CD34+, V+, Bcl-2+	10 months	(50)
Beyer et al, 2012	46	M	RLig	21x7	N	HRT, chemo therapy, resection	CD34+	10 months	(51)
Soussan et al, 2013	64	M	L	N/A	N	Resection	CD34+, Bcl-2+	N/A	(52)
Liu et al, 2013	42	M	L	1.5x1x1	N	Resection	CD34+, Bcl-2+	N/A	(53)
Jakob et al, 2013	62	F	L	N/A	N	Resection	CD34+, CD99+, Bcl-2+	N/A	(54)
Debs et al, 2014	65	M	L	N/A	N	Resection	CD34+, CD99+, Bcl-2+	12 months	(55)
	87	F	R	14.6x12.3x17	N	Nil	N/A	10 months	
Guray-Durak et al, 2013	38	F	L	8x6x2	N	Resection	CD34+, CD99+, SMA+	N/A	(56)
Vythianathan and Jim, 2013	78	M	L	17x13	N	Resection	CD34+, CD99+, V+, Bcl-2+	N/A	(57)
Song et al, 2014	49	M	L, R	7.6x5x4.8	N	Resection	CD34+, V+, Bcl-2+	3 months	(58)

Table I. Continued.

First author, year	Age, years	Sex	Lobe	Size, cm	Hypo	Treatment	IHC	Follow-up	(Refs.)
Texeira *et al*, 2014	68	F	L	7.5x6.5x5.5	N	Resection	CD34+, V+	28 months	(59)
Du *et al*, 2015	55	F	L	11x17x15	Y	Resection	CD34+, Bcl-2+	60 months	(60)
Beltran, 2015	58	M	L	15x9x6	N	Resection	CD34+, V+	36 months	(61)
Bejarano-Gonzalez *et al*, 2015	79	F	R	15	N	TACE, resection	CD34+, V+, Bcl-2+	31 months	(62)
Feng *et al*, 2015	51	M	R	2.3x0.3	N	Resection	CD34+, Bcl-2+	11 months	(63)
	49	M	L	8.7	N	Resection	CD34+, V+, Bcl-2+	17 months	
	51	F	R	8.4	N	Resection, adjuvant chemotherapy	CD34+, V+, Bcl-2+	31 months	
	52	F	R	12	N	Resection, MWA	CD34+, V+	37 months	
Silvanto *et al*, 2015	65	M	L	18	N	Resection	CD34+, CD99+, Bcl-2+	16 months	(64)
Kueht *et al*, 2015	40	M	L	4.7x4x4	N	Resection	CD34+, CD99+, V+, Bcl-2+	N/A	(65)
Maccio *et al*, 2015	74	F	R	24x16	N	Resection	CD34+, V+, Bcl-2+, STAT6+	Succumbed after 15 months	(66)
	80	F	R	19x15	N	Chemotherapy	CD34+, V+, Bcl-2+, STAT6+	Succumbed after 4 months	
	65	M	R	3x2	N	Chemotherapy	CD34+, V+, Bcl-2+, STAT6+	Succumbed after 5 months	
Makino *et al*, 2015	55	M	R	8.6x6.3	N	Resection	CD34+, CD99+, Bcl-2+	11 months	(67)
Dey *et al*, 2016	56	F	R	23x22x10	N	Resection	CD34+, V+, Bcl-2+	6 months	(68)
Degnan *et al*, 2016	39	M	L	20x18x15	Y	Resection	CD34+, V+, Bcl-2+, STAT6+	6 months	(69)
Macak *et al*, 2016	64	N/A	R	20x14.6x19	Y	Resection, TACE	NSE+, CD34+, V+, Bcl-2+, STAT6+	N/A	(70)
Chen and Slater, 2017	61	M	R	15x11.5x7.5	N	Resection	CD34+, CD99+, Bcl-2+	74 months	(71)
Present case	32	F	L, R	19x19.6x14	N	Ex situ hepatectomy and liver autotransplantation	CD34+, CD117+, Caldesmon+, SMA+, PR+	3 month	-

Hypo, hypoglycemia; IHC, immunohistochemistry; N/A, not available; F, female; M, male; L, left; R, right; RLig, round ligament; TACE, transarterial chemoembolization; HRT, hormone replacement therapy; MWA, microwave ablation; CD, cluster of differentiation; V vimentin; Bcl-2, B-cell lymphoma 2; STAT6, signal transducer and activator of transcription 6; NSE, neuron specific enolase; SMA, smooth muscle actin; PR, progestrone receptor.

The *ex situ* hepatectomy and liver autotransplantation was initially performed by Pichlmayr *et al* (4) in 1988 for treating liver tumors involving major vascular structures. The procedure has progressed and developed over the past 30 years, and has been successfully applied for the treatment of several types of lesions, including hepatocellular carcinoma (72,73), hilar cholangiocarcinoma (74), giant hepatic hemangiomas (75) and metastatic colon cancer (72,73,76). The reported cases are listed in Table II. The median follow-up and survival times for these cases were 13 and

Table II. Data on reported ex situ liver autotransplantation cases.

First author, year	Age, years	Sex	Diagnosis	Characteristics of lesion/contraindication to traditional resection	Follow-up	(Refs.)
Pichlmayr et al, 1988	40	N/A	Metastatic leiomyosarcoma	Bilateral liver metastases	N/A	(4)
Yagyu et al, 1994	N/A	N/A	Intrahepatic cholangiocarcinoma	Involved the confluence of the 3 main hepatic veins and the retrohepatic IVC	No recurrence at 8 months	(78)
Hemming and Cattral, 1999	50	M	Colorectal liver metastases	A single lesion in the caudate lobe involved the origin of all 3 hepatic veins	N/A	(76)
Lodge et al, 2000	55	F	Colorectal liver metastases	Involvement of IVC and segments 1, 2, 3, 4a and 8	Post-operative chemotherapy, 30 months survival	(79)
			Colorectal liver metastases	Involvement of segments 2 and 4-8	Succumbed from complications of renal and respiratory failure on post-operative day 15	
			Colorectal liver metastases	17x15 cm mass involving segments 1, 2 and 4-8	Alive at 5 months	
			Colorectal liver metastases	17x13 cm mass involving segments 1, 2, 4-8	Alive at 5 months	
Oldhafer et al, 2000	40	F	Metastatic leiomyosarcoma	Involvement of segments 2, 3 and 5-8	Succumbed at 36 months due to tumor recurrence	(72)
	46	F	Metastatic colon cancer	Involvement of segments 5-7	Succumbed at 13 months due to tumor recurrence	
	52	M	Klatskin's tumor	Required right hepatic trisegmentectomy	Succumbed at 13 months due to tumor recurrence	
	58	M	Metastatic colon cancer	Metastatic colon cancer infiltrating the IVC	Succumbed at 44 days due to sepsis and hepatic insufficiency	
	30	F	Focal nodular hyperplasia	Large FNH in segment 4 with compression	Alive at 9 years of the IVC	
	57	M	Metastatic colon cancer	Infiltration of RHV requiring extended left hemihepatectomy	Succumbed at 21 months due to tumor recurrence	
	48	F	Klatskin's tumor	Klatskin's tumor requiring extended left hemihepatectomy	Succumbed on post-operative day 50 due to sepsis	
	62	M	Klatskin's tumor	Tumor invading left and right portal veins	Succumbed on post-operative day 113 due to sepsis	
	55	F	Klatskin's tumor	Klatskin's tumor requiring right hepatic trisegmentectomy	Succumbed on post-operative day 35 due to sepsis	
	35	M	HCC	HCC requiring resection of segments 1-4	Lost to follow-up and wedge 6	
	43	M	HCC	HCC requiring resection of segments 5 and 6	Succumbed at 25 months due to tumor recurrence	

Table II. Continued.

First author, year	Age, years	Sex	Diagnosis	Characteristics of lesion/contraindication to traditional resection	Follow-up	(Refs.)
	67	M	Metastatic colon cancer	Lesions requiring resection of segments 1-4b and wedge 6-7	Succumbed at 2 months due to intracerebral bleed	
	53	M	Metastatic colon cancer	Lesions requiring resection of segments 1 and 4-8	Succumbed at 2 months due to sepsis	
	54	F	Focal nodular hyperplasia	Lesions requiring resection of segments 1, 4, 5 and 7	Alive at 5 years	
	55	M	Metastatic colon cancer	Lesions requiring resection of segments 1 and 4	Succumbed at 15 months due to tumor recurrence	
	55	F	Metastatic leiomyosarcoma	Lesion requiring resection of segments 2-4b and wedge 5	Succumbed at 13 months due to tumor recurrence	
	52	M	Metastatic colon cancer	Lesions requiring resection of segments 1, 5 and 8	Succumbed at 36 months due to tumor recurrence	
	40	F	Metastatic leiomyosarcoma	Lesions requiring resection of segments 1, 4-8 and wedge 2-3	Succumbed at 18 months due to tumor recurrence	
	52	M	Metastatic colon cancer	Lesions requiring resection of segments 1 and partial 4	Succumbed on post-operative day 14 due to pneumonia	
	52	M	Cholangiocarcinoma	Lesion requiring resection of segments 1-5 and wedge 8	Succumbed on post-operative day 41 due to sepsis	
	39	M	HCC	Lesion requiring resection of segments 1-4 and partial 8	Succumbed on post-operative day 23 due to sepsis	
	71	M	Metastatic colon cancer	Lesions requiring resection of segments 1 and 5-8	Alive after 13 months	
Chui *et al*, 2003	26	F	Cholangiocarcinoma	2.3-cm hilar mass involving the portal vein confluence and right hepatic artery	Alive with no recurrence on MRI at 17 months	(80)
Gruttadauria *et al*, 2005	41	M	Cholangiocarcinoma	14x12 cm mass encompassing segments 4a, 4b and 5 with RHA involvement	Recurrence at 10 months and 13 months, receiving radiation; still alive at 23 months	(81)
	58	M	Leiomyosarcoma of the IVC	Third attempt at resection	Discharged on post-operative day 10; long-term outcome N/A	
Ikegami *et al*, 2007	39	F	Giant hepatic hemangiomas	4 large hemangiomas in segments 1-3, caudal 4 and 6-7	Normal liver function with a regenerated liver graft at 8 months	(75)
Sugimachi *et al*, 2010	17	F	HCC	18x12 cm lesion involving IVC, RHV and first left branch of portal vein	Alive 28 months after resection; no post-operative chemotherapy	(82)
Gringeri *et al*, 2012	38	F	Hepatic metastasis from pancreatoblastoma	2.5 cm lesion in left lobe and 2.7-cm lesion in right lobe involving MHV and RHV	Alive after 8 months	(83)
Zhang *et al*, 2012	60	F	Hepatic hemangiomas	Lesions requiring resection of segments 4-8	Alive at 22 months	(74)
	64	M	Cholangiocarcinoma	Lesions requiring resection of segments 1-4	Alive at 17 months	
	55	M	Cholangiocarcinoma	Lesions requiring resection of segments 1, 5, 7 and 8	Succumbed on post-operative day 1 due to liver and renal failure	

Table II. Continued.

First author, year	Age, years	Sex	Diagnosis	Characteristics of lesion/contraindication to traditional resection	Follow-up	(Refs.)
Wen et al, 2013	67	M	HCC	18x12 cm lesion at the confluence of the LHV, MHV and IVC	Alive at 28 months, no recurrence	(73)
	71	M	HCC	18x13 cm lesion at the confluence of the V7/RHV into the IVC	Alive at 26 months, recurrence treated with RFA	
	60	M	HCC	5.8x6.8 cm lesion centrally located, involving the RHV and PV	Alive at 23 months, recurrence treated with TACE	
Baker et al, 2015	66	F	Extensive cholangiocarcinoma	Lesions involving all 3 hepatic veins	Alive at 3 months	(6)
Vicente et al, 2017	51	M	Cholangiocarcinoma	Lesions involving retrohepatic vena cava together with the 3 hepatic veins	Alive at 36 months	(77)
Present case	32	F	Solitary fibrous tumor	Lesions involving the right and left hepatic arteries	Alive at 3 month	N/A

F, female; M, male; FNH, focal nodular hyperplasia; HCC, hepatocellular carcinoma; IVC, inferior vena cava; LHV, left hepatic vein; MHV, middle hepatic vein; PV, portal vein; RFA, radiofrequency ablation; RHV, right hepatic vein; RHA, right hepatic artery; TACE, transarterial chemoembolization; V7, hepatic vein branch draining segment VII of the liver.

25 months, respectively. In addition, 63.6% (28/44) patients receiving *ex situ* hepatectomy and liver autotransplantation were <55 years old, as was the patient reported in the present case. The common indications for this method are major vein involvement and extensive resection area, both of which were noted in the present case. Compared with conventional liver resection, *ex situ* hepatectomy and liver autotransplantation markedly improve the rate of successful resection and clear margins of the otherwise unresectable lesions. There is no long waiting time for a suitable allograft, as an autologous liver is implanted. In addition, the total cost is lower and no post-operative immunosuppressants are required for patients receiving *ex situ* hepatectomy and liver autotransplantation, compared with an allograft liver transplantation (5).

The successful establishment of venous reflux and hypothermic perfusion of the isolated liver are essential for securing a successful surgery and a good prognosis (77). As the liver resection and trimming surgery are time-consuming, the prolonged anhepatic phase can lead to hemodynamic or internal environmental disturbances. Therefore, in the present case, an artificial blood vessel was used to establish an end-to-side portocaval shunt in order to maintain the venous return of the intestinal tract and lower limbs during the anhepatic period. Compared with autologous vessels, including the great saphenous vein, use of an artificial blood vessel minimizes the invasiveness of the procedure for the patient. As the shunt is temporary, the potential tissue rejection is of no concern.

The commonly encountered complications of *ex situ* hepatectomy and liver autotransplantation include bile leakage, bleeding, pulmonary infection, pneumonedema, hydrothorax and renal insufficiency, with a total incidence of 58.1%. The overall mortality rate within 90 days after surgery is 19.5% (5). In the present study, the patient presented with moderate right pleural effusion following surgery, and recovered fully following pleurocentesis and drainage. No other severe complications were encountered up to 3 months after surgery.

In conclusion, this is the first report that describes the successful adoption of *ex situ* hepatectomy and liver autotransplantation for treating an otherwise unresectable giant SFT. The uneventful surgical course and post-operative period suggested that this procedure was suitable for managing an unresectable liver neoplasm. Furthermore, the artificial blood vessels may provide a reliable source for establishing an end-to-side portocaval shunt during the anhepatic period.

Availability of data and materials

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

Authors' contributions

SY and WW conceived and designed the study. ZS, YD, YJ, QZ, ZL, JX, JD, SY and WW performed the surgery and provided the patient care. QZ, ZL, JX and JD performed the literature review and collected the substantial data. ZS and YJ analyzed the data. ZS and YD wrote and revised the manuscript. ZS, YD and YJ organized the data and figures. SY and WW revised the final version of manuscript. All authors have read and approved the manuscript.

Ethics approval and consent to participate

The study was approved by the Ethics Committee of the First Affiliated Hospital, School of Medicine, Zhejiang University.

Patient consent for publication

The patient provided written informed consent for the publication of any associated data and accompanying images.

Competing interests

The authors declare that they have no competing interests.

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