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Inflammatory myofibroblastic tumor of the liver: A case report and review of literature

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Abstract

BACKGROUND

Inflammatory myofibroblastic tumors of the liver (IMTL) are extremely rare neoplasms and very little is known about their clinical presentation, pathogenesis, and biological behavior. Due to their absolute rarity, it is almost impossible to obtain a definite diagnosis without histological examination. Because of their intermediate biological behavior with the risk for local recurrence and metastases, surgical resection is recommend whenever IMTL is suspect.

CASE SUMMARY

We herein present a case of an otherwise healthy 32-year-old woman who presented with intermittent fever, unclear anemia, malaise and right flank pain 4 mo postpartum. The liver mass in segment IVa/b was highly FDG avid in the positron emission tomography-computed tomography. Hepatic resection was performed achieving a negative resection margin and an immediate resolution of all clinical symptoms. Histological analysis diagnosed the rare finding of an inflammatory myofibroblastic tumor of the liver and revealed cytoplasmic anaplastic lymphoma kinase expression by immunohistochemistry. Twelve months follow-up magnetic resonance imaging showed no recurrence and no metastases in the fully recovered patient.

CONCLUSION

IMTLs are extremely rare and difficult to diagnose. Due to their intermediate biological behavior, surgical resection should be perform whenever feasible and patients should be followed-up in order to detect recurrence and metastasis as early as possible.

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Core tip: In summary of the literature and with the experience from our own recent case, complete surgical resection of suspected inflammatory myofibroblastic tumors of the liver should be the preferred treatment of choice in order to rule out malignancy, avoid long-term medical treatment and to be able to recommend an appropriate follow-up for the patient.

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INTRODUCTION

Inflammatory myofibroblastic tumors (IMT) are rare diagnostic findings and little is known about their etiology, pathogenesis and clinical behavior. First described in the lungs, this rare neoplasm can occur in various tissues and organs of the human body^[1-4]. Whereas IMTs were originally considered as inflammatory pseudo tumors, they are now recognized as true neoplasms in the histological typing of the soft tissue tumors classification of the World Health Organization with intermediate biological potential due to their ability to recur and to metastasize^[1,4]. IMTs of the liver (IMTL) are even more seldom and most published literature are case reports (Table 1) or small case series (Table 2). Most patients present with either abdominal pain or fever, in others the tumor is detected incidentally^[5]. A systemic inflammatory process with leukocytosis, elevated C-reactive protein (CRP) and other systemic inflammatory markers often accompanies the clinical presentation^[3,5-11]. Although this type of neoplasm can occur in individuals of all ages, it seems more common in children and young adults^[4,12]. The etiology of IMTL is unclear^[4], but cytogenetic alterations suggest a clonal origin of these lesions^[3,4]. Proof of diagnosis is difficult since no tumor markers are available and radiological findings are often not specific^[6,8,13]. Surgical resection is usually considered as the treatment of choice for these rare findings. IMTLs mostly present as solitary lesions with typical firm surfaces. Histopathologically, they can have three basic patterns, which are often combined in one tumor: (1) A myxoid/vascular; (2) Spindel cell; and (3) Hypocellular fibrous pattern^[4]. The tumor is frequently infiltrated by eosinophils, lymphocytes and plasma cells^[4]. Rearrangements of the anaplastic lymphoma kinase (ALK) gene locus are common in IMTs supporting its neoplastic origin. ALK overexpression and its positive immunohistochemical staining is reported in 50%-60% of the cases^[14]. Differential diagnoses of IMTL include metastatic sarcomatoid carcinoma, spindel cell sarcoma or melanoma, gastrointestinal stromal tumor, sarcoma, solitary fibrous tumor and calcifying tumors besides the large group of inflammatory pseudotumors^[6]. Although these lesions generally show a benign behavior, there is the possibility of malignant transformation and development of metastases^[15,16]. Some small case series of IMTs described the anatomic location, size and age as potential risk factors correlated with recurrence^[2,15,17]. In addition, ALK reactivity in the primary tumor was associated with a non-metastatic course of the disease^[6]. In the liver, a malignant transformation is extremely rare and only very few cases with local recurrence or metastases have been described^[1,18]. Due to the scarcity of this disease, the role of a preoperative biopsy is unclear, but because of the difficulty to obtain a proper histopathological diagnosis and the risk of malignant transformation, surgical resection is usually recommended whenever technically feasible^[5,8,9,19,20].

We herein report the case of a 32-year-old woman who received an immediate hepatic resection for a large IMTL causing intermittent fever 4 mo postpartum.

CASE PRESENTATION

Chief complaints

A 32-year old woman presented herself to her family doctor with intermittent fever, unclear blood loss, malaise and pain in the right flank 4 mo postpartum.

History of present illness

The patient reported that the symptoms began 4 mo after she gave birth to her healthy child. She complained about fatigue and right upper quadrant abdominal pain. She had recurrent episodes of fever up to 38.5 °C, but no jaundice or pruritus.

History of past illness

There was no significant history of past illnesses.

Personal and family history

Personal and family history was unremarkable. She gave birth to a healthy child 4 mo before she was treated at our institution.

Physical examination upon admission

Vital signs were within the normal range, body temperature was 38.5 °C. On examination, the patient had a right upper quadrant tenderness, without jaundice or hepatosplenomegaly.

Laboratory examinations

Urine and most blood analyses were without any pathological findings including a normal liver function and normal ferritin levels. While the white blood cell count was normal, CRP was elevated to 181 mg/L. The liver enzymes (aspartate-aminotransferase 31 U/L, alkaline-aminotransferase 49 U/L) and cholestasis parameters (alkaline-phosphatase 466 U/L, γ -glutamyl transferase 424 U/L) showed an increase while the serum bilirubin (6 μ mol/L) stayed normal.

Imaging examinations

An ultrasound of the abdomen (**Figure 1A**) revealed a round, encapsulated liver lesion in segment IVa/b of unclear dignity, a non-contrast computed tomography of the abdomen ruled out urolithiasis, but confirmed the suspicious lesion of 8 cm in the liver as an incidental finding. The computer tomography (CT) and, same day magnetic resonance imaging (MRI) of the upper abdomen (**Figure 1B-F**) showed an 8 cm \times 8 cm tumor in segment IVa/b of the liver suspected to be a liver adenoma. Additional serological tests for hepatitis, the tumor markers carbohydrate-antigen 19-9 and alpha-fetoprotein, and markers for echinococcosis were all negative. After discussion of the case in our interdisciplinary liver tumor board on the next day, we performed a positron emission tomography-computed tomography (PET-CT) which showed the known lesion as a metabolically active tumor resembling an inflammatory pseudotumor of the liver or a malignant tumor of unclear origin. No other lesions were detected in any of the performed scans.

Further diagnostic work-up

The pathologist macroscopically (**Figure 2**) described the size of the resected specimen as wedge-shaped and nodular, 9.5 cm \times 7.0 cm \times 7.5 cm. The capsule of the liver was about unremarkable on one-half of the specimen. An area of 7.5 cm \times 7.5 cm \times 6.2 cm was sharply circumscribed, whitish/creamy and fibrous. No clearly definable capsule. The remaining liver tissue was inconspicuous and showed no further hereditary findings. The total weight of the tumor was 198 g. Immunohistochemistry showed a clear expression of cytoplasmic ALK and a weak expression of smooth muscle actin. Cytokeratin-PAN (CK Pan), Cytokeratin 18 (CK18), signal transducer and activator of transcription protein 6 (STAT6), Desmin, tyrosin-protein (C-kit), discovered on gastrointestinal stromal tumors 1 (DOG1), ETS related gene (ERG), family of calcium binding protein (S100) and SRY-related HMG-box 10 Protein (SOX10) showed no expression. The intra-tumoral immunoglobulin G4 (IgG4)-positive plasma cells were slightly increased, but displayed only a very small percentage of all plasma cells (**Figure 3**). The pathological diagnosis revealed an IMTL with no fibrosis and no malignancy.

FINAL DIAGNOSIS

The final diagnosis of the presented case is an IMTL.

Table 1 Case reports

Ref.	n	Age (yr)	Gender	Clinical and laboratory findings	Radiology	Localization	Tentative diagnosis	Treatment	Histology	Follow up
Watanabe <i>et al</i> ^[23] , 2019	1	70	Female	Incidental finding	CT unenhanced, low density	Right lobe	HCC	Right partial hepatectomy	Unencapsuled, partly ill defined expansive mass, myofibroblast-, fibroblast after 7 mo cells, inflammatory cells, SMA+, cytokeratins AE1/AE3+; CK7,CK18+, Desmin-, CD68-, IgG4+, ALK-	No recurrence
Al-Hussaini <i>et al</i> ^[24] , 2019	1	8	Male	FUO, weight loss, hepatomegaly, normal liver enzymes, CRP↑	MRI: Contrast-enhancing, hyper-intense, well-defined lesion	Right lobe	Infection DD malignancy	Right lobe hepatectomy	Multinucleated giant cells, inflammatory cells, SMA-, ALK-1-, CD-21- CD-23- CD-68+	No recurrence after 4 mo
Lu <i>et al</i> ^[25] , 2018	1	20	Male	FUO, jaundice, abdominal pain, CA 19-9↑	MRI: Multiple lesions, intrahepatic bile duct was significantly dilated	Left lobe	CCC	Biopsy, patient declined operation, PTCD	Spindle cells proliferation and infiltration by mixed inflammatory cells, ALK+, SMA+	NM
Jin <i>et al</i> ^[6] , 2017	1	42	Female	Fatigue, fever, pale conjunctivae; Hb↓, Lc↑	U/S: Hypochoic mass with unclear border; CT: Low density lesion with mild enhancement	Right lobe	Liver abscess	Right posterior segmentectomy	Chronic inflammatory cells, spindle cells; CD68+, smooth muscle actin, ALK-	No recurrence after 32 mo
Muлки <i>et al</i> ^[26] , 2015	1	50	Male	Abdominal pain, anorexia, mild fever, hepatomegaly	U/S: 2 hypodense masses, CT: + hepatic vein thrombus	Right lobe	Abscess with septic thrombus	Initial treatment: Biopsy, pigtail, antibiotics, secondary operation	Plasma cells, inflammatory cells, ALK, IgG4+	No residual disease
Obana <i>et al</i> ^[25] , 2015	1	69	Male	FUO, CA 19-9 48 ng/mL (<i>ti</i> : < 37 ng/mL), Diabetes mellitus II, Dyslipidemia, hypertension	U/S: Irregularly shaped, low-echoic mass; CT: Peripherally enhanced, MRI: T1W, central portion hyperintense	Right lobe Seg VI	CCC/HCC	Partial hepatectomy	Whitish-yellow mass 2 cm in size, inflammatory cell infiltrates, cholesterol cleft granuloma with focal abscess were observed in the central compartment, IgG4 -	NM
Guerrero Puente <i>et al</i> ^[24] , 2015	1	75	Male	Weight loss, fever, intermittent night sweat, abdominal pain, CRP↑, leukocytosis, cholestasis hypertension, hypercholesterinemia	CT: 8 cm heterogeneous focal lesion, portal branch thrombosis, lymphadenopathy; MRI: T2W isointense, T1W discretely hypointense, cystic-necrotic areas, perilestional edema	Left lobe	Inflammatory disease	CT-guided biopsy followed by antibiotic therapy	Inflammatory pseudotumour, vimentin+, AML+, desmin-, CD68-, ALK-, with no light chain restriction and a low proliferative index (15%)	Partial remission after 1 mo, almost complete remission after 6 mo
Onieva-González <i>et al</i> ^[27] , 2015	1	70	Male	Low-grade fever, asthenia, weight loss and oligoarthritis, lung tuberculosis, diabetes, gouty arthritis, renal lithiasis and colon diverticulitis	CT: Thickened gallbladder wall, poorly-defined hypodense lesion of 17 mm in the gallbladder bed, U/S: Nodule; MRI: Hypointense in T2 sequences, PET: No metabolism	Seg. V	Liver abscess	Antibiotic therapy, after 4 mo later fine needle biopsy followed by laparoscopic biopsy and cholecystectomy with the lesion in the gallbladder bed	Lymphoid infiltration without malignancy signs, compatible with an inflammatory pseudotumour	NM
Chang <i>et al</i> ^[28] , 2014	1	38	Male	Fatigue, abdominal distension and weight loss, jaundice, hepatomegaly, bilateral ankle edema	U/S: Complex mass; CT: Large cystic or necrotic mass; MRI: T2W: Cystic portion hyperintense to liver parenchyma, surrounded by a hypointense rim. T2W: Hyperintense compared to liver parenchyma	Bilateral	N/A	Ultrasound-guided and open biopsy, followed by resection	Cellular spindle-cell proliferation with heavy inflammatory infiltrate consisting primarily of plasma cells and lymphocytes	Recurrence after 2.5 yr

You <i>et al.</i> ^[33] , 2014	1	43	Male	Chronic cough, right-upper-quadrant pain, anorexia for 3 mo, leukozytosis, elevated platelet count	U/S: 18 cm mass with slightly echogenic center; MRI: Large mass with central dark area and some peripheral spokes; CT: Mass, 20 cm × 17 cm × 18 cm, with extensions into the medial segment of the left hepatic lobe, hypervascular nodular area with enhanced density at the periphery and hypoaattenuating density centrally	Right lobe	Fibrolamellar hepatocellular carcinoma or CCC	Percutaneous needle core biopsy > NM	Bland spindle cell proliferation amidst small mature lymphocytes, numerous plasma cells, histiocytes, and few neutrophils. Spindle cells showed a storiform pattern with large areas of necrosis; cytokeratin (CAM5.2), cytokeratin 5/6-, actin-, CD34-, CD117-, DOG-1-, desmin-, CD68-, S100-, Pan-melanoma-. Spindle cells were negative for CD21, CD23, CD35, ALK-1. Epstein-Barr virus-encoded small RNA in situ hybridization (EBER) showed large numbers of Epstein-Barr virus positive cells, including some spindle cells	NM
Durmus <i>et al.</i> ^[34] , 2014	1	67	Female	Moderate diffuse abdominal tenderness, focus over epigastrium	U/S: Heterogeneous hypoechoic tumor; CT: Contrast enhancing mass with irregular confluent non-enhancing areas in the center with a hypodense late enhancing rim and no wash-out in the late phase, MRI: In T1W hypointense borders, well defined without fatty components. T2W showed a heterogeneous slightly hyperintense lesion with an ill-defined hyperintense rim	Segment IV	Malignancy	Left hemihepatectomy with partial excision of the adherent abdominal wall and diaphragm	Tumor with fibrosis and partially necrotic tissue infiltrated by inflammatory cells, predominantly plasma cells, and also pigmented macrophages and granulocytes	NM
Wong <i>et al.</i> ^[37] , 2013	1	56	Female	Right-upper-quadrant abdominal pain, renal transplant	U/S: 2 cm × 2.4 cm mass in the left hepatic lobe with associated biliary duct dilatation, MRI: atrophic left liver lobe with multiple strictures and distal duct dilatation. 2-cm lesion at the origin of the left hepatic duct	Left lobe	Primary hepatic tumor	Surgical resection	Dense hyalinised stroma and scattered, histiocytic and lymphocytic inflammation	NM
Kruth <i>et al.</i> ^[38] , 2012	1	NM	NM	FUO CRP↑	Gastroscopy, CT lung and abdomen, MRI: 3.3 cm lesion	Seg. VI	Adenoma, focal nodular hyperplasia or HCC	Surgical resection	NM	No recurrence after 1 yr
Chablé-Montero <i>et al.</i> ^[39] , 2012	1	23	Female	Fever, diaphoresis, right-upper-quadrant abdominal pain	U/S and CT: Heterogeneous rounded hepatic lesion of 7 cm in greatest dimension	Right lobe	Pyogenic hepatic abscess	Antibiotics, later right hepatic lobectomy	Grossly a non-encapsulated but well demarcated hepatic tumor with central necrosis of 11 cm in greatest dimension microscopically: Spindle myofibroblastic cells arranged in fascicles. Leukocytes, lymphocytes, plasma cells, SMA+	NM

Kayashima <i>et al</i> ^[30] , 2011	57	Female	Asymptomatic laparoscopic calculous cholecystectomy 3 yr ago	U/S: 3 liver masses; CT: 1 intra- and 2 extrahepatic lesions; MRI: three high-intensity lesions; PET: Abnormal accumulation in all lesions	Right lobe	CCC	Surgical resection (tiny black-colored nodules within the abdominal cavity and spilled gallstones)	Inflammatory granuloma located at liver parenchyma	No recurrence after 6 mo
Huang <i>et al</i> ^[40] , 2012	30	Male	Right upper abdominal pain; CEA↑; 2 yr after renal transplant	CT: Low-density mass, about 30 mm in diameter, well defined, and with peripheral enhancement CT: Numerous hypodense lesions scattered throughout the liver U/S: Ill-defined area, CT: multiple low attenuation lesions	Caudate lobe	HCC or liver abscess	Hepatic caudate lobectomy with complete resection of the mass Liver biopsy	Mixture of spindle-shaped myofibroblastic cells and chronic inflammatory cells; SMA+	NM
Beauchamp <i>et al</i> ^[41] , 2011	74	Female	FUO	lesions scattered throughout the liver	NM	NM	Liver biopsy	IMT	NM
Al-Jabri <i>et al</i> ^[31] , 2010	69	Male	Right upper quadrant pain, nausea, vomiting, recent weight loss, rheumatoid arthritis and bronchiectasis, CRP↑, cholestasis (normal Bilirubin)	U/S: Ill-defined area, CT: multiple low attenuation lesions	Right lobe	Cholecystitis, malignancy	Fine needle biopsy	Presence of benign hepatocytes, acellular debris and a mixture of acute and chronic inflammatory cells	No recurrence after 3 mo
Salakos <i>et al</i> ^[43] , 2010	10	Male	Fever, weight loss, fatigue, tachycardia, hepatomegaly, leukocytosis, platelet count ↑	U/S: Space occupying lesion in the liver; CT: Large lesion with solid and cystic parts and heterogeneous enhancement	Right and left lobe	NM	Biopsy followed by conservative treatment (ceftriaxone, clindamycin, NSAR)	Hyperplastic cholangioles, myofibroblasts and fibroblasts, infiltrate of lymphocytes, eosinophils and neutrophils; ALK+ response	Partial response after 2 mo, complete response after 18 mo
Ueda <i>et al</i> ^[45] , 2009	79	Male	Leukocytosis	U/S: Hypoechoic lesion, 3 cm in diameter, with several stones. CT: Low density area in segment V; MRI: Lesion of slightly low signal intensity; MRCP: Lesion of moderate-to-high signal intensity on T2W	Right lobe	Inflammation due to cholangitis with intrahepatic bile duct stones	1. ERCP: Sphincterotomy, antibiotics because of common bile duct stone. 2. Relapse of symptoms 4 wk later > resection	Grossly gray, fibrotic, solid tumor, intrahepatic bile duct stones. Proliferation of diffuse myofibroblastic and mesenchymal cells in a mixed myxoedematous, dense fibrotic stroma, with many small vessels and marked infiltration by various acute and chronic inflammatory cells	No recurrence after 18 mo
Stirer <i>et al</i> ^[7] , 2009	48	Female	Weakness, fever, weight loss, right upper abdominal pain, Leu-, neutrophil 75.3%, liver function normal	U/S: Single hypoechoic lesion in right lobe	Right lobe	NM	Resection	No capsule, light brown, no necrosis, spindle cells, granulation-tissue type vessels, chronic inflammatory cells on loose, edematous, myxoid stroma, CD38+, SMA+, ALK+, desmin, EMA-	2 yr no recurrence after 2 yr
Manolaki <i>et al</i> ^[47] , 2009	9	Female	Fever, mild anorexia, intermittent epigastric pain	U/S: Hypoechoic lesion, lymph node at porta hepatis, CT: hypodense space-occupying lesion	Left lobe	NM	Biopsy, secondary left lateral segmentectomy with lymph node excision	Pale and firm lesion (3.5 cm x 2.5 cm x 3.0 cm) with whitish solid infiltrations extending to the capsule of the liver. Proliferation of spindle-shaped cells arranged in short fascicles with an ill-defined mark. Inflammatory cells, predominantly lymphocytes, plasma cells and eosinophils; vimentin+, SMA+, CD68+, JBC+	No recurrence after 3 yr

CT: Computed tomography; MRI: Magnetic resonance imaging; FUO: Fever unknown origin; CRP: C-reactive protein; CCC: Cholangiocarcinoma; HCC: Hepatocellular carcinoma; PTCD: Percutaneous transhepatic cholangio drainage; NM: Not mentioned; U/S: Ultrasonography; Hb: Haemoglobin; L.C: Leukocytes; TC: Thrombocytes; TIW: T1-Weighted; T2W: T2-Weighted; Chron Hep B: Chronic Hepatitis B; Seg: Segment; ↑: Increase; ↓: Decrease; WBC: Wight blood cells; SMA: Smooth muscle actin; ERCP: Endoscopic retrograde cholangiopancreatography.

Table 2 Clinical studies of > 2 patients

Ref.	n	Age (yr)	Gender	Clinical and laboratory findings	Radiology	Localization	Tentative diagnosis	Treatment	Histology	Follow up
Park <i>et al.</i> ^[28] , 2014	45	65 (29-84)	Male/female (26/19)	Abdominal pain (n = 16), fever (n = 11), malaise (n = 5), weight loss (n = 4); CRP↑ (n = 31), leukocytosis (n = 10), CEA (n = 1) CA 19-9 (n = 1); hypertension, tuberculosis, chronic Hepatitis B	CT scan: Hypo-attenuating lesions in 40 patients, MRI: Low signal intensity lesion at T1W image in 86.4% and relatively homogenous high signal intensity lesion at T2W image in 76.2%	Right lobe (n = 27), left lobe (n = 14), both (n = 4)	Malignancy (n = 26, 57.8%), abscess (n = 11, 24.4%)	Percutaneous needle biopsy (n = 35), surgical resection (n = 9), both (n = 1)	Chronic infiltration of various inflammatory cells (plasma cells, lymphocytes, neutrophils, and eosinophils) and fibrous stroma	No recurrence after median follow-up of 8 mo
Ahn <i>et al.</i> ^[29] , 2011	22	34-76	Male/female (16/6)	Abdominal pain (n = 12), febrile (n = 5), malaise (n = 1), asymptomatic (n = 4), leukocytosis (n = 6), hyperbilirubinaemia (n = 3), alkaline phosphatase↑ (n = 10), liver enzymes ↑ (n = 5), CA 19-9 ↑ (n = 5), AFP↑ (n = 1); associated biliary disease (n = 15), malignancy (n = 4)	Solitary (n = 17); multiple (n = 5), median size 3 cm (1.1-9.6 cm), non-enhanced CT: Hypoattenuating lesions (n = 22), enhanced CT: Central hypoattenuating areas and a delayed hyperattenuating periphery (n = 18), multiseptate appearance with hyperattenuating internal septa and periphery (n = 3), hypoattenuation up to the equilibrium phase (n = 1)	Right lobe n = 10, left lobe n = 9, both n = 3, (mostly seg. IV n = 12)	IPT (n = 12), malignancy (n = 4), abscess (n = 4)	Percutaneous needle biopsy (n = 18), incisional biopsy (n = 1) --> surgical resection (n = 3); liver resection (n = 3) without prior biopsy, 16 patients conservatively, 6 patients with surgical resection	Histiocytic cell infiltration with negative IgG4 (n = 17), lymphoplasmacytic type (n = 5) with positive IgG4 (n = 4)	Post conservative treatment: 10 complete remission after 15 mo; 5 partial remission after 4 mo, post resection: Mortality n = 2 (myocardial infarction, peritoneal seeding)
Geramizadeh <i>et al.</i> ^[44] , 2009	2	14	Male	Chills, fever, anorexia > 8 kg weight, leukocytosis	CT: Well-defined heterogeneous mass with central areas of necrosis and a slightly hyperdense rim	Left lobe	Abscess	Resection	Creamy grey mass with a vague whorling appearance. Plasma cells with varying degrees of fibroblastic proliferation admixed with lymphocytes, eosinophils and macrophages	No recurrence after 1 yr
Yamaguchi <i>et al.</i> ^[27] , 2007	3	52	Male	Hepatitis B positive, weight loss, Epigastric pain, appetite loss, weight loss, fever	Well defined liver mass	NM	Malignancy	Fine needle biopsy	6 cm liver mass, fibroblastic proliferation, many plasma cells and eosinophils	No recurrence after 2 yr
		58	Male	Auxiliary finding	CT: Low density mass in the right lobe enhanced during the delayed phase	Right lobe	CCC	Biopsy > no treatment, follow up	IMTL	NM
		57	Female	Sigmoid cancer planned for resection	MRI: 2 metastases with low-intensity signal on T1, a slightly high-intensity signal on T2	Right lobe	Hepatic metastasis	Intraoperative right portal vein embolization	NM	NM

Author	Year	Age	Sex	Presenting symptoms	Imaging findings	Pathology	Management	Outcome
Millas <i>et al</i> ^[10]	2009	35	Male	Abdominal and bone pain, fatigue, malaise, hematuria, WBC [↑]	CT: Liver abscess right upper abdominal quadrant	Right lobe	Drainage followed by right hepatectomy	Many plasma cells, densely collagenous bundles between a plasma cell-rich infiltrate
		56	Male	Right upper abdominal pain, malaise	CT: Liver abscess	Right lobe	Drainage followed by right hepatectomy	Inflammatory response to hepatic abscess
		75	Female	Moderate upper quadrant pain, nausea, and vomiting	U/S: Cystic lesion, CT: Cystic lesion, slight dilatation of intrahepatic bile ducts	IVB	Biopsy followed by Seg, IVB resection	Central granulation, fibrosis and chronic lymphoplasmacytic infiltrate, no features of neoplasia.
		47	Female	Right upper quadrant pain, jaundice, fever, pruritus	CT: Marked dilatation of the intrahepatic biliary tree	Right lobe	Seg. III resection, secondary right hepatectomy	Inflammatory pseudotumor Widespread chronic inflammatory infiltrate with lymphocytes and plasma cells, numerous lipid-laden macrophages, no malignancy

CT: Computed tomography; MRI: Magnetic resonance imaging; F/UO: Fever unknown origin; CRP: C-reactive protein; CCC: Cholangiocarcinoma; HCC: Hepatocellular carcinoma; PTCD: Percutaneous transhepatic cholangio drainage; NM: Not mentioned; U/S: Ultrasonography; Hb: Haemoglobin; LC: Leukocytes; TC: Thrombocytes; TIW: T1-Weighted; T2W: T2-Weighted; Chron Hep B: Chronic Hepatitis B; Seg: Segment; ↑: Increase; ↓: Decrease; WBC : Wight blood cells; SMA: Smooth muscle actin; ERCP: Endoscopic retrograde cholangiopancreatography.

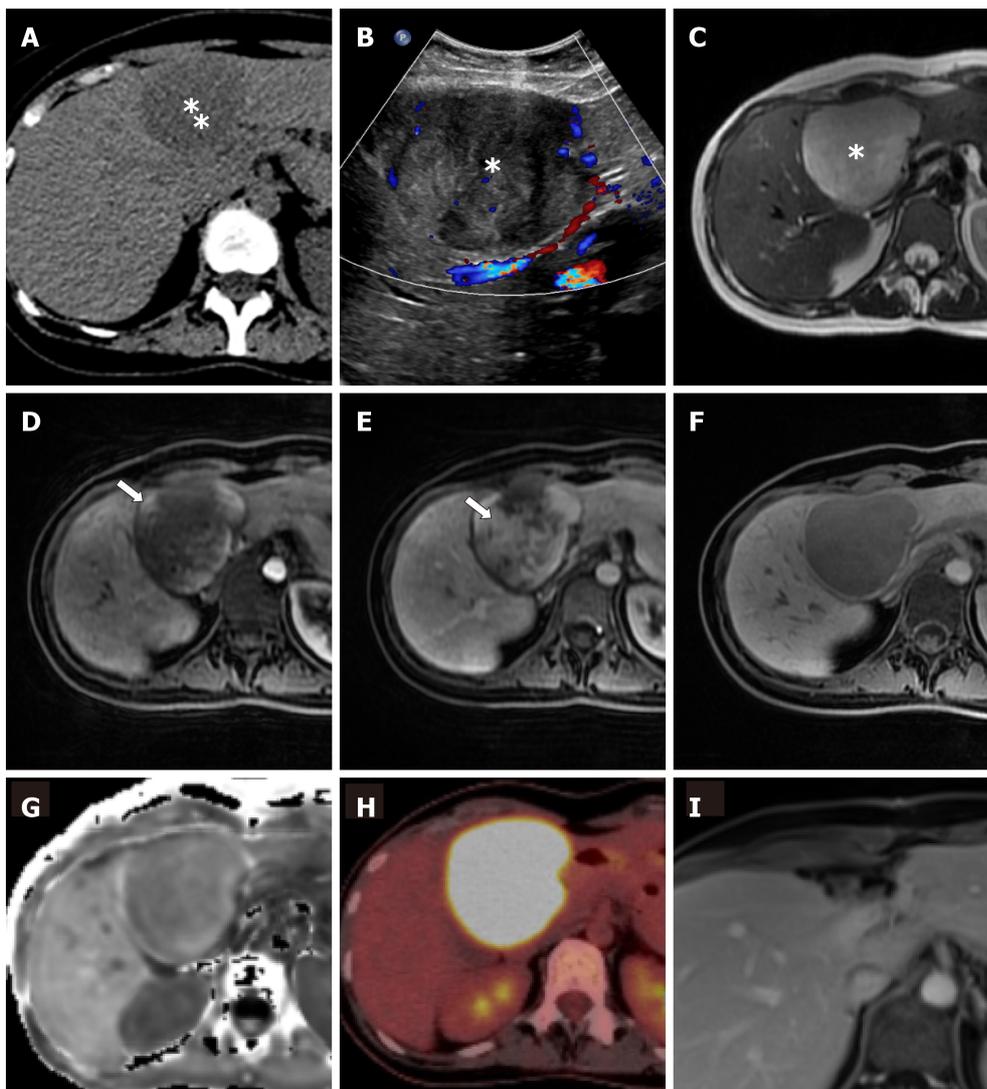


Figure 1 Imaging features within the liver lesion in segment IV. A: The lesion was a first detected as an incidental finding in an unenhanced abdominal computed tomography to rule out kidney stones (asterisk); B: Conformed with an ultrasound examination (asterisk); C: In a following magnetic resonance imaging the lesion showed a homogeneous high signal in T2-weighted imaging (asterisk); D: After the application of intravenous hepatocyte specific contrast medium (gadoteric acid, Primovist®/Eovist®, Bayer Healthcare Pharmaceuticals, Leverkusen, Germany) there was an early enhancement at the rim in the arterial phase (arrow); E: Followed by a strong enhancement in the venous phase (arrow); F: In the hepatobiliary phase after 20 min, the lesion appeared with a low intracellular uptake of the contrast medium compared with the adjacent liver tissue; G: In the diffusion-weighted imaging there was no clear diffusion restriction detection within the lesion (apparent diffusion coefficient); H: In an additional positron emission tomography-computed tomography examination the lesion showed an intensively increased tracer uptake; I: A follow-up magnetic resonance imaging examination after 3 mo confirmed a complete surgical resection (with multiple artifacts at the resection margin due to multiple clips) and ruled out new hepatic lesions.

TREATMENT

Due to the unclear situation with fever and the suspicion of a large adenoma or malignant tumor of the liver, an immediate surgical resection was performed. Intraoperatively, the solitary central lesion could be confirmed by intraoperative ultrasound, which also excluded additional liver lesions. An open resection of the liver segment IVa/b was performed achieving a negative resection margin. While no intra-operative complications occurred, the patient developed a bilioma, which had to be drained interventionally 7 d after the surgery accompanied by an endoscopic retrograde cholangiopancreatography with stent insertion.

OUTCOME AND FOLLOW-UP

The case was discussed postoperatively in our interdisciplinary liver tumor board to determine the postoperative management. While no adjuvant therapy was indicated, it was recommended to follow the patient clinically by MRI imaging every 3 mo after



Figure 2 Postoperative macroscopic pathology of the inflammatory myofibroblastic tumors.

the surgery for the duration of at least one year.

The patient returned to work and MRIs of the liver 3, 6 and 12 mo after resection showed no local recurrence and no novel liver lesions.

DISCUSSION

We herein present and discuss the case of a 32-year-old woman who presented with a suspicious and symptomatic liver mass consequently diagnosed as IMTL.

IMTs of the liver are extremely rare findings that can sometimes mimic malignant lesions^[6]. In terms of demographics, the tumor seems to be more common in men than in women (M/F: 1.5/1) with a mean age at diagnosis of 37 years^[7]. IMTL usually occur in the right liver lobe, in close proximity to the gallbladder or central biliary system^[7,8]. Typical clinical findings reported in the literature are fever, abdominal pain, lack of strength and weight loss^[7], which all occurred in our case (intermittent fever, unclear blood loss, malaise and pain in the right flank) and led to the ultimate diagnosis. In addition to the fever, laboratory findings often suggest inflammation due to leukocytosis, neutrophilia and elevated CRP^[5,6,8,10]. More rarely, anemia and sometimes also elevated liver enzymes are reported^[9]. According to the clinical signs of infection, some individual cases were reported to be correlated with different active (virus) infections^[5,18,19,21,22]. In our patient, the antibody to Epstein-Barr virus was positive in the serological findings without any signs of an active Epstein-Barr virus infection. A clear association between IMT and infectious organisms seems to be doubtful since in most reported series, including our own case, no acid-fast organisms, fungi, parasites or bacteria could be identified in the tumor^[10,19].

Radiological features of IMTLs are nonspecific and a definite radiological diagnosis seems to be impossible. Due to the small cases (Tables 1 and 2) we could see, that the tumor in ultrasonography mostly was hypoechogenic. An IMT may be suspected if a defined soft tissue mass and a heterogeneous enhancement with invasive or non-invasive growth are present on adjacent structures in CT or MRI^[6,8,23]. Not all patients underwent a MRI for diagnostic treatment, only in eight cases^[17,24-29]. Al-Hussaini *et al.*^[24] and Kayashima *et al.*^[30] described a contrast-enhancing, hyper-intense well defined lesion without going into details. In four cases the lesion in T1W was mostly hypointense and T2W hyperintense^[17,25,26,28]. Despite its rarity, lack of diagnostic signs and symptoms, IMTL should not be ruled out as a differential diagnosis in liver lesions like focal nodular hyperplasia, hepatocellular adenoma, carcinoma and echinococcosis especially in young patients with normal tumor markers^[7]. In addition IMTL can sometimes mimic a liver abscess^[22]. Although many synonyms have been used for this lesion, including plasma cell granuloma, postinflammatory tumor, xanthomatous pseudotumor, inflammatory pseudotumor, and inflammatory fibrosarcoma^[31], the new classification clearly suggests the term inflammatory myofibroblastic tumor of its suitable origin or organ, in our case an IMTL^[4].

Due to the small number of cases worldwide (Tables 1 and 2), no clear diagnostic tests or radiographic features exist that help to make a definite diagnosis without a histopathological examination of the tissue^[10]. We performed a comprehensive literature search and studied the cases published during the last 10 years^[5,7,17,24-30,32-47]. There were more men affected than women. The most common localization of the tumor was on the right lobe of the liver. All patients in the described cases had at least an ultrasonography and/or a CT. In some cases, the diagnostic work-up was completed with MRI, MRCP or PET-CT. Due to the different radiological findings the

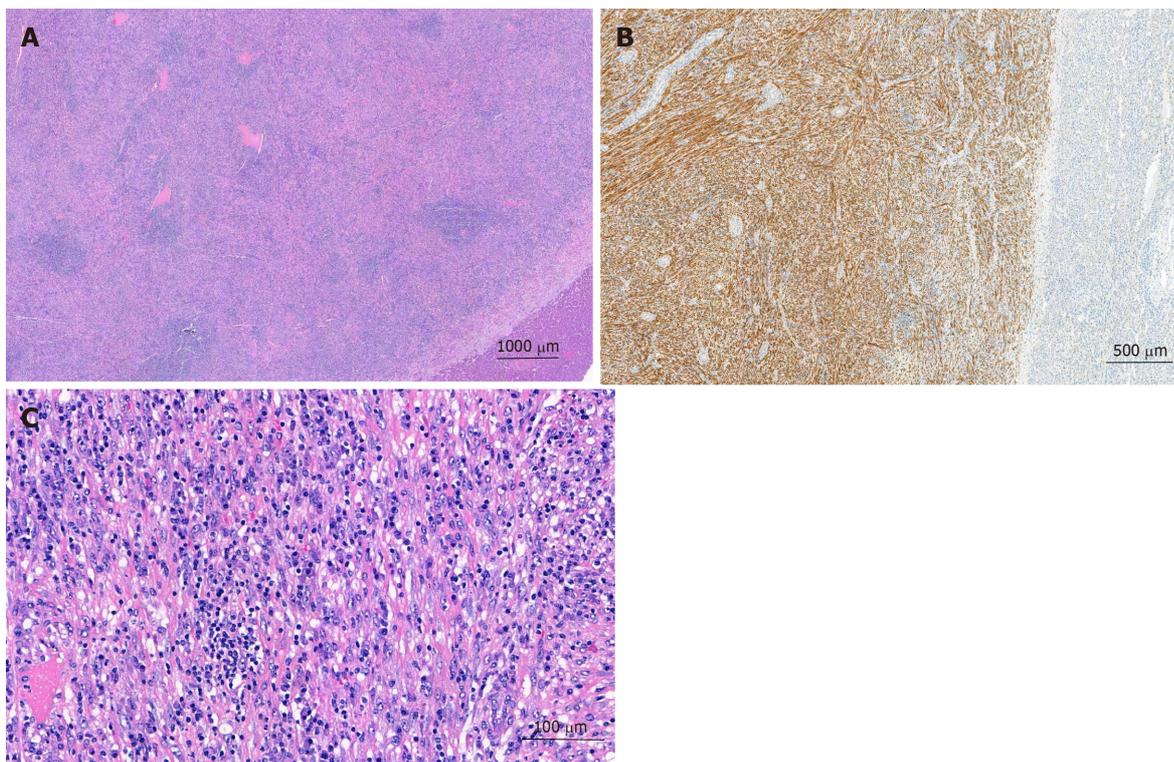


Figure 3 Postoperative microscopic pathology of the inflammatory myofibroblastic tumors. A: Well demarcated firm vascularized tumor mass with spotty inflammatory infiltrate; B: Bland proliferation of spindle cells in broad fascicles at higher magnification. Scattered lymphocytes and plasma cell; C: Intense positivity of the spindle cells for anaplastic lymphoma kinase.

tentative diagnose showed a large variation from liver abscess, inflammatory process and also malignancy.

In the gross examination of the resected specimen, most findings showed the similar finding of a well-demarcated, unencapsulated, yellow-whitish mass. Histologically infiltrations of chronic inflammatory-cells like lymphocytes, neutrophils, eosinophils, and macrophages were often described. Whenever immunohistochemical analyses were performed, ALK expression showed a similar distribution. The performed treatment of the different cases varied according to the initially suspected diagnose. In summary, more patients were treated conservatively, although there is no clear indication for such a treatment. Surgical resections were performed according to the size and location of the suspected tumor and varied from small atypical resections to major hepatectomies. In most of the cases the definite histology report of the resected specimen then showed the diagnosis of an IMTL. Unfortunately, follow-up was not described in all published cases. Except for one reported recurrence after 2.5 years, most patients stayed tumor-free during a follow-up ranging from X-Y months^[48].

Surgical resection is usually recommended so that a proper pathological work-up can be performed and malignancy can be ruled out. Nevertheless, several different treatment strategies have been published including conservative approaches with steroids, high-dose steroids, radiation and chemotherapy^[6-8,11]. Interestingly, one case with a spontaneous regression has also been reported^[17]. A typical pathological finding is that the IMTL's are unencapsulated. They are usually solid or gelatinous on the intersection and have a white color. Hemorrhage, calcification or necrosis are rarely described^[6,12], similar to the pathological findings in our case. As described by Elpek *et al*^[6], chromosomal translocations leading to the activation of ALK can be detected in IMTLs. Although immunohistochemistry for ALK expression in immunohistochemistry can reliably predict the presence of ALK gene rearrangement, its prognostic relevance is still unclear^[14,49]. IMTLs differ from IgG4-related liver disease in terms of ALK expression, low IgG4 positive cell infiltration, and lack of obstructive phlebitis^[6].

The natural course of IMTL without curative surgical therapy is unclear. To date, only a few cases have been described in which patients had local recurrence or metastases after liver resections^[15,16,48]. Due to the small numbers published worldwide, no recommendations for the follow-up are available and patients is treated according to the decisions made in the local interdisciplinary tumor boards. In

our case, the finding of the pseudotumor was 4 mo postpartum. Due to the rather large size of the lesion it was considered an advanced lesion. The pregnancy may have masked general symptoms such as nausea, vomiting, and abdominal pain. So far, only one case of newly diagnosed IMTL has been reported during pregnancy^[18].

CONCLUSION

In summary of the literature and with the experience from our own recent case, complete surgical resection of a suspected IMTL should be the preferred treatment of choice in order to rule out malignancy, avoid long-term medical treatment and to be able to recommend an appropriate follow-up for the patient.

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