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Int J Hepatobiliary Pancreat Dis 2023;13(1):16–25.
www.ijhpd.com

CASE REPORT PEER REVIEWED | OPEN ACCESS

A unique presentation of peliosis hepatis: A case report and comprehensive review of the literature

Alex Teshon, Crystal Walker, David P Schammel, Christine Marie-Gilligan Schammel, A Michael Devane

ABSTRACT

Introduction: Peliosis hepatis (PH) is a rare benign vascular condition characterized by dilatation of hepatic sinusoids with occasional involvement of other organs. While associated with chronic immunosuppression, anabolic steroid use, oral contraceptive (OCP) use, human immunodeficiency virus/acquired immunodeficiency syndrome (HIV/AIDS), and infection with Bartonella, most PH patients are asymptomatic and, thus, identified incidentally. Compromised liver function is sometimes evident through laboratory tests; however, mortality results from cyst rupture and hemorrhage spontaneously or during surgical procedures.

Case Report: We report a case of PH identified in a 33-year-old Black female radiologically evaluated for abnormal liver function tests. Computed tomography (CT) revealed enhancement of >100 lesions throughout both liver lobes; a CT-guided biopsy revealed mild macrovesicular steatosis and marked sinusoidal dilation, consistent with peliosis hepatis.

Conclusion: We also present a comprehensive literature review describing the associated conditions, pathology, diagnostic methods, and treatment options for PH patients.

Keywords: Benign vascular liver condition, Dilation of sinusoidal liver spaces, Peliosis hepatis

Alex Teshon[1], BS, Crystal Walker[2], BS, David P Schammel[3], MD, Christine Marie-Gilligan Schammel[3], PhD, A Michael Devane[2], MD

Affiliations: [1]University of South Carolina School of Medicine Greenville, Greenville, SC, USA; [2]Department of Radiology, Prisma Health Upstate, Greenville, SC, USA; [3]Pathology Associates, 8 Memorial Medical Ct., Greenville, SC, USA.

Corresponding Author: Christine Marie-Gilligan Schammel PhD, Director, Clinical Research, Department of Pathology, Prisma Health Upstate, SC, USA; Pathology Associates, 8 Memorial Medical Ct., Greenville, SC 29605, USA; Email: Christine.schammel@prismahealth.org

Received: 15 March 2023
Accepted: 08 May 2023
Published: 31 May 2023

How to cite this article

Teshon A, Walker C, Schammel DP, Schammel CMG, Devane AM. A unique presentation of peliosis hepatis: A case report and comprehensive review of the literature. Int J Hepatobiliary Pancreat Dis 2023;13(1):16–25.

Article ID: 100103Z04AT2023

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doi: 10.5348/100103Z04AT2023CR

INTRODUCTION

Peliosis hepatis (PH) is a rare benign vascular condition with a reported incidence between 0.2 and 22%, characterized by dilatation of hepatic sinusoids and occasional involvement of other organs [1–6]. Risk factors include chronic immunosuppression, anabolic steroid use, oral contraceptive (OCP) use, HIV/AIDS, and infection with *Bartonella* spp [2, 4, 7–13]. Most cases are discovered incidentally on imaging or during routine follow-ups for abnormal lab results [2, 9, 14–18]. Peliosis hepatis lesions may spontaneously or surgically rupture and hemorrhage, which is associated with significant mortality [4, 8, 19–22]. Interestingly, reports note spontaneous regression of lesions with cessation of causative medications, normalizing liver function tests [6, 13, 23]. Peliosis hepatis lesions can be identified by sonography, CT, magnetic resonance imaging (MRI), and angiography. However, the definitive diagnosis is determined by the pathologic pattern of disease, stages of the blood component in the lesions, and hepatic steatosis for differentiation from hepatic adenomas,

hemangiomas, hepatic abscesses, or hepatocellular carcinoma [16]. Treatment for persistent PH is resection to avoid progression to cholestasis, portal hypertension [1, 4, 12], and hepatic failure; however, biopsies and resection are associated with significant bleeding risk and mortality [1, 9, 24, 25].

Following Institutional Review Board (IRB) approval and consent, we present radiographic findings of PH in an asymptomatic patient with stable lesions successfully managed with regular radiographic follow-up. Additionally, we present a comprehensive review of the literature describing the pathology, associated conditions, diagnostic methods, and treatment options for PH.

CASE REPORT

A 33-year-old black female with a past medical history of seizures, hypertension, type-2 diabetes, cognitive impairment, and neuropathy resulting in complete loss of ambulation was referred to our institution for radiologic evaluation following an abnormal liver function test (alkaline phosphatase (ALP) 1193; alanine transaminase/aspartate transaminase (ALT/AST) were normal at 16/16 U/L), microcytic anemia, and a vitamin B12 deficiency. No alcohol, drug, tobacco use, or family history of liver disease were noted. On physical exam, jaundice and abdominal pain were absent; chronic fatigue had been present for several months. Abdominal ultrasound (Figure 1A and B) revealed hepatomegaly that exceeded 23 cm, with multiple heterogenous masses in both lobes of the liver hyperechoic to the surrounding parenchyma; further evaluation with contrast-enhanced CT (Figure 2A–C) revealed enhancement of approximately 100 lesions throughout both liver lobes, the largest measuring 5–6 cm. The liver parenchyma had a uniform density without pathologic enhancement and a normal gallbladder without evidence of biliary dilation. Magnetic resonance imaging (MRI) (Figure 3A–C) shows numerous T2 hyperintense hepatic lesions, which enhanced during the arterial phase with intravenous (IV) gadolinium. To rule out a hyper-vascular primary malignancy or vascular metastasis, a CT-guided percutaneous biopsy of the liver was performed. Pathology identified mild macro-vesicular steatosis and marked sinusoidal dilation (Figure 4A–C), consistent with peliosis hepatis and mild macro-vesicular steatosis of the liver. Occult bleeding of the upper and lower gastrointestinal tract (GI) tract was ruled out; the anemia was treated with Vitamin B12 injections and IV iron transfusions. After two years of follow-up, the anemia persisted; testing revealed alfa-thalassemia minor. Radiographic follow-ups noted stable lesions; the patient has remained asymptomatic three-years post-diagnosis. Interestingly, however, the patient's lab values have continued to show elevated ALP levels that vary between 1200 and 1700 IU/L, while the ALT/AST values have remained normal.

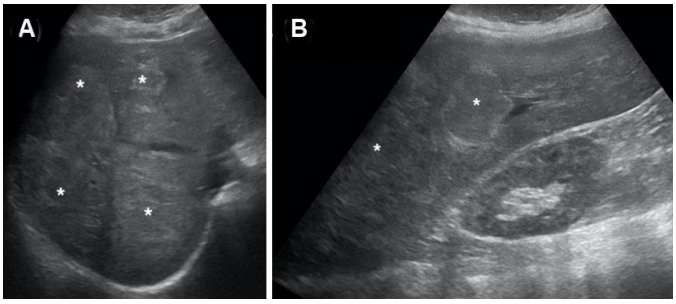

Figure 1: (A) Transverse and (B) longitudinal ultrasound images of the liver demonstrate hyperechoic lesions (*) scattered throughout the liver.

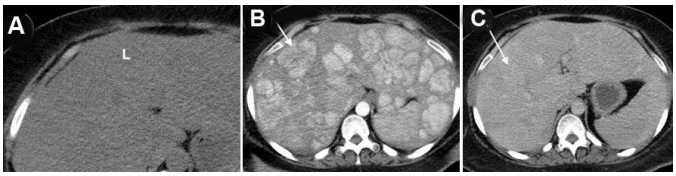

Figure 2: (A) Non-contrast, (B) arterial phase, and (C) portal venous phase intravenous contrast enhanced computed tomography of the abdomen shows multiple enhancing liver lesions in the arterial phase (one example noted with white arrows) that are not clearly delineated without intravenous contrast (A) and less conspicuous in the venous phase (A). L=liver, S=spleen.

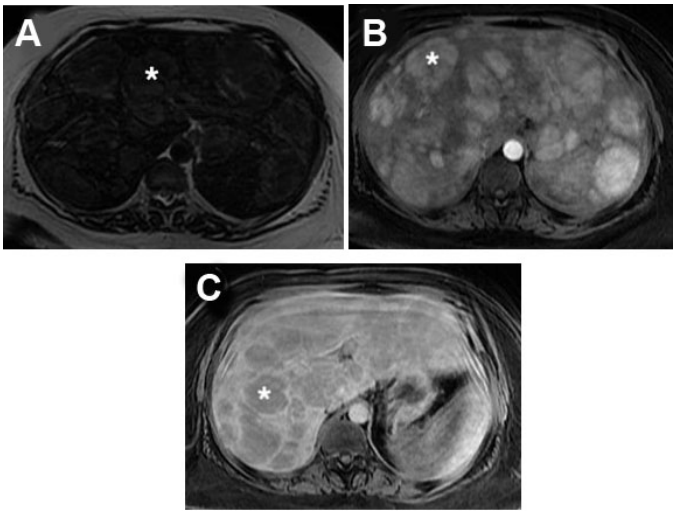

Figure 3: Magnetic resonance imaging of the liver. (A) T2 sequence demonstrates numerous hepatic lesions (*) that are mildyT2 hyperintense. (B) These lesions (*) enhance avidly in the arterial phase with intravenous gadolinium. (C) Liver lesions (*) are relatively hypointense in the venous phase sequence.

DISCUSSION

Peliosis hepatis has been associated with certain medications, infections, hematologic conditions, and transplantation [1, 4, 7, 13, 26–29] (Tables 1 and 2). Symptomatic PH (n=49; 78%) presents with abdominal

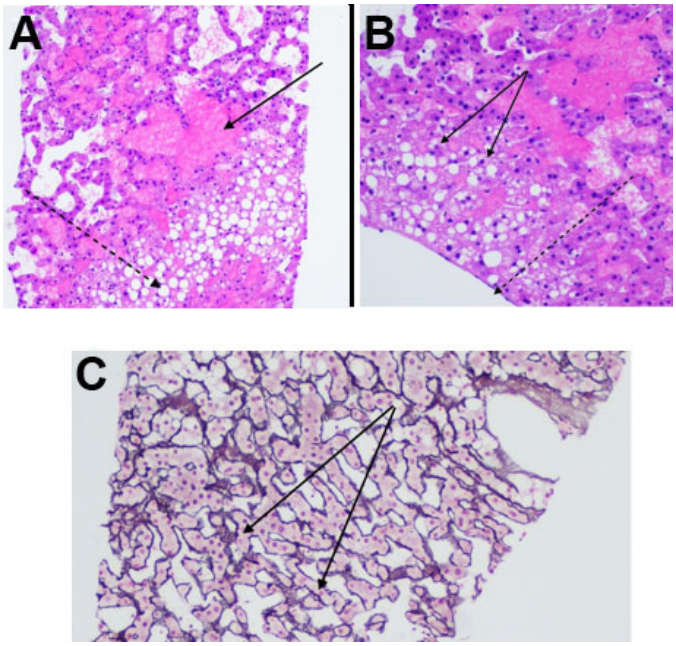

Figure 4: Histology (A) hepatic dilated sinusoids filled with blood (solid arrow) and attenuated hepatocellular cords; fat noted as well (broken arrow; H&E staining; 100×). (B) Higher power highlighting hepatocytes intermingled with fat (solid arrow) and a vein (broken arrow; H&E staining; 200×). (C) Reticulin stain highlighting the hepatocellular cords (solid arrow; 100×).

pain (40%; n=25), GI symptoms/weight loss (21%; n=13), and abdominal fullness/hepatomegaly/mass (19%; n=12; Table 1) [1, 3, 4, 6, 30]. Fatigue, as in our patient, was reported rarely (8%; n=5). Of those that reported outcomes (n=48), mortality was 27% (n=13).

Abnormal lab values (64%; n=40), specifically, elevated liver function tests (38%; n=24), were common [1, 3, 9, 31]: ALP (52.6%; n=10), ALT (47.4%; n=9), gamma-glutamyl transferase (GGT) (42.1%; n=8) and AST (31.6%; n=6). Interestingly, the elevated ALP in our patient (1100–1700 IU/L) is the highest reported (mean 391 IU/L; Table 1), and the consistently normal AST/ALT is the first reported (Table 1) [1, 6, 12]. Anemia (35%; n=22), thrombocytopenia (19%; n=12), leukocytosis (11%; n=7), and hyperbilirubinemia (10%; n=6) were also reported. For patients with anemia, as with our patient, colonoscopy (7.9%; n=5) and upper GI endoscopy (EGD) (15.9%; n=10) were completed (Table 1).

A majority of asymptomatic patients (22%; n=14; Table 1) were diagnosed incidentally by imaging (71%; n=10), with variable findings within lesional tissue and the concomitant presence of hepatic steatosis [9, 16, 17, 26, 32]. Vascular tumors, hepatic adenomas, hepatocellular carcinoma, hepatic abscesses and other cystic liver diseases remained in the differential [4, 6, 12, 13, 18, 27], with normal magnetic resonance pancreatography (MRCP) (6.3%; n=4) and fluorodeoxyglucose-positron emission tomography (FDG-PET) (11.1%; n=7) ruling out malignant processes [1, 4, 12].

Diagnostic imaging is essential in evaluating PH due to biopsy/surgery bleeding risk. Initial ultrasound (55.6%; n=35) identified homogenous/hypoechoic lesions with hepatic steatosis or hyperechoic lesions with normal hepatic parenchyma [16, 26], as noted in our patient (Figure 1). Doppler studies can identify blood flow and thrombi or portal obstruction [6, 16, 26]. Contrast-enhanced CT (54.0%; n=34), the preferred imaging modality [4, 16, 18, 26, 32–34], differentiates hypoattenuating peliotic lesions versus normal parenchyma [16, 18, 32–35]; characteristically, early central enhancement during the arterial phase with centrally accumulating contrast in a "target sign" that diffuses to the periphery during the venous phase is noted in PH [16, 18, 26, 33–35]. With an active hemorrhage, hyperattenuating contrast material may accumulate or leak during the late phase [4, 16, 18, 26, 32–34]. The initial CT in our report revealed multiple enhancing lesions throughout the liver parenchyma, the largest 5–6 cm (Figure 2).

Magnetic resonance imaging (MRI) was a commonly utilized imaging modality (40.0%; n=25) with MRI T1-weighted images appearing hypointense with an enhancement that progresses centrifugally; cavities display a rim of enhancement with hematoma [17, 18, 26, 35]. T2-weighted MRI images appear hyperintense to the parenchyma with multiple foci of high signal, consistent with what was seen in our patient using Gadolinium contrast enhancement (Figure 3) [16–18, 26].

Diagnostic angiography identifies contrast accumulation during the late arterial phase and persistent enhancement during the venous phase with multiple vascular nodules [4, 6, 8, 20, 36]; angiography also allows embolization to commence [36–38]. Interestingly, reported angiography noted a right-sided predominance of PH (70.6%) with a greater incidence of hemorrhage, most often from the right hepatic artery (62.5%).

Definitive diagnosis of PH currently relies on histologic identification of blood-filled cystic dilated sinusoids throughout the liver parenchyma with evidence of rupture of reticulin fibers or necrosis of hepatocytes and macrovesicular steatosis (Figure 4) [4, 6, 10, 12, 15, 27, 30, 39]; however, due to the risk of hemorrhage (26.3%; n=12) or death (41.7%; n=5), biopsy is only warranted when imaging reveals a concern for malignancy [6, 12, 25, 39]. Thus, monitoring of PH through serial imaging and liver function testing [1, 11, 40–43], may be appropriate.

Prophylactic surgical resection of affected tissue may be considered in patients with worsening cavitary lesions to prevent spontaneous cyst rupture and hemorrhage (n=7) [4, 8, 19, 20–22], but this can be complicated by hemorrhage and death (n=1; Table 1) [44]. Emergent surgical management or embolization is reserved for those with evidence of acute hemorrhage (36.5%; n=23) [6, 8, 36, 40]: surgical resection (56.5%; n=13), embolization (39.1%; n=9), and embolization followed by surgical resection (n=1). Complications and mortality

were noted following resection (23%; n=3 and 30%; n=1, respectively) and embolization (44.4%; n=4 and 50%; n=2, respectively), but reports are few.

Currently, there is no specific treatment for PH, and lesions have been reported to spontaneously resolve without clinical consequence (7.9%; n=5) [4, 13, 23, 29]; specifically, drug/toxicity-related cases (7.9%; n=5) often resolve with withdrawal of the offending drug/toxin with normalization of lab values. Peliosis hepatis lesions due to Bartonella may regress with antibiotics, such as Doxycycline or Erythromycin; however, bacillary peliosis cases were not identified in the literature [45]. Typically, however, progression to portal hypertension and liver failure occurs when PH remains untreated (22%; n=14) [1, 4, 6, 13, 29] with ascites (58.3%; n=7), portal hypertension (25%; n=3), liver failure (25%; n=3), and encephalopathy (8.3%; n=1) reported; mortality due to long-term complications of hepatic failure was also noted (16.7%; n=2).

While there is no consensus for routine monitoring in asymptomatic patients or those who refuse surgery, serial imaging (55.6%; n=5) and liver function tests have been suggested (14.3%; n=9) [37]; intervals of three months (n=3), six months (n=2), and one year (n=1) have been reported. Of these, 44.4% had stable disease, regression/normal labs were noted in 33.3% and 22.2% died from progressive hepatic failure.

Table 1: Comprehensive literature review: Case reports (n=63)*

Author	Year	Age	Sex	Symptoms	Labs	US	CT	MRI	Other imaging	Histology	Tx	Outcomes	Associated conditions
Ferrozzi	2000	35	F	RUQ pain, HM	Nl	BS, HS, multiple HoL	HS, multiple HrL b/a contrast	Hemorrhagic collection T1HI, T2HI	GdCT	HS, CBS w/o EL		Increase lesion size, subcapsular blood collection	OCP use
Gouya	2001	50	F	Abd fullness, WL	Nl	HoL in RHL, Nml USD	HoL,	T1HI, T2HI		CBS w/o necrosis		Lesions stable	Estrogen HRT
Shim	2001	33	M	Liver mass		No focal lesions	HrL in RHL in P phase	T1HI, T2HI	Hepatic angiography	CBS w/ EL	Resection		HBV
Verswijvel	2003	74	M	Abnormal LFTs		HoL w/ cystic spaces in RHL		T1HI, T2HI	GdCT	CBS		Regression of lesion size	Steroid therapy
Verswijvel	2003	68	F	Flank pain		HyL mass in RHL		T1HI, T2HI cystic and slit-like PEL		HN, CBS w/o EL			Breast cancer, tamoxifen
Corpa	2004	72	M	DOE, LE edema, WL	MA				Roentgenogram	CBS, lymphoid infiltrate, CD20+		Upper GI bleeding, HS, death	Lymphoplasmacytic lymphoma
Hiorns	2004	3	F	Abd pain, fever	PCTP, DIC	HM, HoL, no flow on USD, cystic fluid	RHL mass, HP, IVC compression					Recovery, Nml appearing liver	
Hyodo	2004	50	M	HM, Abd distension, ascites						CBS	Liver transpl		Liver cirrhosis
Karger	2004	30 months	M	Autopsy						CBS w/o EL		HH, death	Spiro-Shy-Gonatas syndrome, gonadorelin therapy
Omori	2004	31	F	RUQ pain, vaginal bleeding		HP, HoL in RHL	HP, HoL in RHL	T1HI, T2HI	Trans-femoral arteriography	Purple spots on the liver	Embol	Discharge	2 months post-partum
Berzigotti	2006	36	M	Abnormal labs	TCP, elevated GGT, LFTs	Coarse liver, Nl flow on USD, portal HTN			BMB, EGD	Periportal fibrosis, obliteration of PV, FNH	Ursodeoxycholic acid	Nml LFTs	Chronic liver dx, portal HTN
Saritas	2006	21	M	HM						CBS	Resection	Resolution of anemia	Castleman dx
Atila	2007	43	M	R pleural effusion, RUQ mass		Large cystic lesion in RHL				CBS w/ EL	R lobectomy	Cyst rupture, death from organ failure	
Elsing	2007	36	M	Fatigue, pruritis, ascites	PCTP GGT 121 ALP 220	HM w/ macronodular surface				Sinusoidal ectasia, portal fibrosis	D/c AZA	Resolution of lesions	Crohn's dx, AZA, steroids
Kim	2007	42	F	RUQ pain, fever	Elevated LFTs, MA, LCT	SCL in RHL w/ fluid	HoL in RHL, heterogenous HrL		Angiography	RHL hematoma, CBS	R lobectomy	Discharge, Nml CT exams	

Table 1: *(Continued)*

Author	Year	Age	Sex	Symptoms	Labs	US	CT	MRI	Other imaging	Histology	Tx	Outcomes	Associated conditions
Tsirigotis	2007	29	M	IF on US	TCP	HoL	Bilateral hyper-vascular lesions, large SCH		Angiography		D/c Danazol, hepatic artery embol	Resolution of lesion	PNH, danazol, allogenic BMT
Tsirigotis	2007	11	F	HM, abnormal LFTs	PCTP, HyB		Bilateral liver lesions,				BMT, d/c oxymetholone	Nml LFTs, resolution of lesions	Fanconi anemia, oxymetholone
Chen	2008	44	F	RUQ pain	Nl	HoL in RHL	HoL w/o CEL			CBS, Syphilitic Gummatous lesion	R lobectomy		
Choi	2009	20	M	RUQ pain, seizures, hypotension	MA, TCP ALT 495 AST 332		Laceration in RHL, hemorrhage		Angiography	HN, CBS w/o EL	R hepatic artery embol, R lobectomy	Discharge	Aplastic anemia, anabolic steroids
Hasimoto	2009	73	M			HrL w/ halo and mosaic pattern		Irregular PEL	Hepatic arteriography	HCC, CBS	Resection		HCC
Kleger	2009	25	F	Fatigue, elevated LFTs WL, anemia	MA GGT 113 ALP 512	HSM, nml echogenicity, nml perfusion to PV	Nml	HSM w/o masses	EGD	Diffusely enlarge hepatic sinusoids	CRT	Resolution of anemia, LFTs	OCPs, Hodgkin's lymphoma
Mungan	2009	36	F	Ascites	MA, HyIG	Nl flow on USD	PV aneurysm		Angiography, EGD	CBS w/ EL	PVS	Death due to sepsis	Portal HTN, intestinal lymphangiectasia
Battal	2010	10	M	Abnormal LFTs, HM		Multiple solid hepatic lesions, HoL		T2HI, w/o T1HI					Fanconi anemia, oxymetholone
Suzuki	2011	60	F	RUQ pain, distension	LCT, MA		Hematoma, RHL lesion, large SCL		Angiography, ERCP, FDG-PET		Embol	HS, resolution of hematoma	
Terasako	2012	58	F	Fever, fatigue, odynophagia	PCTP, ALT 2400, AST 6800	HoL	Multiple HoL w/ CEL			HN, Cowdry bodies	Plasmapheresis	HF, death	Aplastic anemia, cyclosporine, methenolone, allogenic HSCT
Xiong	2012	75	F	RUQ mass	HyB		HoL in RHL	T1HI, T2HI, P/D phase PEL		CBS	Resection	None	Colon cancer, capecitabine, oxaliplatin
Alessandrino	2013	51	M	IF on US				T2HI, T1HI, D phase CEL					Hereditary Hemorrhagic telangiectasia
Dai	2013	19	M	RUQ pain	HyB	Neoplasm in RHL	PEL in A phase			CBS w/o EL, HN	Resection	Discharge	
Gronlykke	2013	33	F	Epigastric pain, N/V	ALP 367 ALT 152 GGT 390	Gallstones, multiple HoL	Early PEL in A phase		CT-US, MRCP	CBS			2 months post-partum, OCP
Motoki	2013	5	M	PNA, abd distension; atelectasis	LCT, MA, TCP		HM w/ heterogenous HoL in RHL, contrast leak during A phase		Hepatic angiogram	CBS w/o EL, HN	Common hepatic artery embol	HH, death	X-linked myotubular myopathy
Pan	2013	38	M	Liver lesion	MA	Nml-appearing liver, mass in RHL		Mass w/ A phase PEL		CBS w/o EL	Resection		
Terlizzi	2013	8	M	Found down	LCT, MA, AST 94 ALT 119		SCH LLL, w/ mass infiltration, HP		Hepatic angiography		Hepatic artery embol	Resolution of liver lesion	X-linked myotubular myopathy
Sanz-Canalejas	2013	44	M	Chronic diarrhea, fever	MA, GGT 404 ALP 546		Ring-enhancing HoL		TTE	Sinusoidal ectasia, CBS, non-necrotizing granulomas	Anti-TB regimen, steroid	None	Disseminated TB, AIDS
Sommacale	2013	44	F	Fever, abd pain shock	MA, TCP, ALT 323 AST 412		Nml		Hepatic angiography	HN	Embol	Death from organ failure	
Tuyama	2013	23	M	SBO, ascites	LCT, MA	Portal HTN on USD, liver nodularity			MRCP	CBS	CT-guided drainage, paracentesis	None	Crohn's dx, 6MP

Table 1: *(Continued)*

Author	Year	Age	Sex	Symptoms	Labs	US	CT	MRI	Other imaging	Histology	Tx	Outcomes	Associated conditions
Lui	2014	59	M	IF on US	HyB	Bilobular nodules, ascites		Multiple lesions w/ CEL	Angiography	CBS w/o EL	Embol	HS, enlargement of lesion	Kidney transplant, cyclosporine, Mycophenolate, methylprednisolone, ESRD
Seo	2014	48	F	IF on CT			HoL		FDG-PET	CBS			Rectal cancer, CRT
Seo	2014	34	M	IF on CT			HoL		FDG-PET	CBS			CRT, gastric carcinoma
Yu	2014	46	M	RUQ pain, N/V	MA, elevated LFTs	Bilateral HrL	Multiple HoL before contrast, PEL in A/P phase			CBS w/ EL, CD34+, CD31+, Ki-67+		Portal HTN, massive ascites, recurrent bleeding varices, hepatic encephalopathy, death	Kidney transplant, mycophenolate, tacrolimus, prednisolone
Cimbanassi	2015	53	F	RUQ pain, HR	MA, ALT 70	HP in RHL	Laceration of RHL, SCH			CBS w/ fibrosis	Resection	Hepatic encephalopathy, no post-op complications	Pregnancy, MTHFR, antiphospholipid syndrome, HRT
Crocetti	2015	29	F	Abd pain		HoL in RHL	HoL	T1HI, T2HI		CBS w/o EL, macrophage histiocytes	Resection		
Downes	2015	58	F	Abd pain, N/V, HP, weakness, ascites			SCH on RHL				Ex-Lap	IPH, discharge	Alcohol use, OCP
Hidaka	2015	77	M	IF on US	Nl	HrL	HoL in A phase,	T1HI, T2HI		CBS w/o EL			Prostate cancer, LH releasing hormone, CRT
Kim	2015	64	F	Epigastric pain, WL	MA, TCP		HEL w/ mosaic perfusion pattern	HM		CBS w/ EL	D/c steroids	Recovery	ITP, steroid use
Koote	2015	47	F	Abnormal LFTs, N/V RUQ pain	MA, ALP 299 GGT 120	Multiple HrL	HEL, HoL in V phase		PET-CT	CBS	D/c OCPs	Liver hematoma, regression of lesions	OCP use
Iwata	2016	75	M	IF on US	Nl	Multiple liver masses	Multiple angiomatous lesions throughout liver, PEL		EGD	CBS w/o EL		HF, death	
Biswas	2017	42	M	RUQ pain, elevated LFTs	MA, TCP	HoL	HoL	T2HI nodules, D phase PEL	Hepatic angiography	CBS w/o EL		HP, death	
Butt	2017	20	F	Fever, RUQ pain	MA, TCP, ALP 510 ALT 316 AFP 64.5	Coarse liver texture, ascites	HM w/ HEL, HoL		EGD	CBS w/ fibrosis		HE, SBP, death	Pregnancy
Dai	2017	64	F	Fatigue, anorexia, RUQ pain	TCP, HyB, ALP 792 GGT 607		HSM, multiple HoL		MRCP	CBS	Abx	Resolution of sx	
Dai	2017	47	M	IF on MRI	Nl	Liver calcification		RHL lesion	EGD	CBS	Resection	Discharge	
Tan	2017	45	F	Abnormal LFTs	AFP 29.4 ALP 121 LDH 400		HEL in A phase	T1HI, T2HI		HM, CBS w/o EL	Resection		Multinodular goiter, progestin analog use
Cordeiro	2018	59	F	RUQ pain	TCP, LCT	Multiple HrL		T2HI D phase PEL				Increased lesions, recovery	SLE, prednisone, cyclophosphamide, AZA, dapsone, Mycophenolate
De la Mano	2018	68	F	Anemia, anorexia sweating, WL	MA, TCP	HM, no focal lesions	HM, HEL w/o focal lesions	HM	EGD, PET	CBS w/ fibrosis		Splenectomy, recovery	Non-Hodgkin follicular lymphoma, R-CHOP
Levin	2018	20	M	GI sx, WL					FDG-PET/CT	CBS			

Table 1: *(Continued)*

Author	Year	Age	Sex	Symptoms	Labs	US	CT	MRI	Other imaging	Histology	Tx	Outcomes	Associated conditions
Yesmembetov	2018	21		HM, portal HTN							Ex-Lap	HH	
Dave	2019	62	F	RUQ pain	PCTP		RHL hematoma, scattered hepatic cysts	RHL hematoma, hepatic cysts	Angiography	CBS w/ fibrosis, HN	Embol	HH	Multiple myeloma, HSCT, HLA-B27 autoimmune dx
Funayama	2019	4	M	Autopsy	AST 2633 ALT 1576		HoL in LLL			CBS, HN, HS		HS, IPH, death	X-linked myotubular myopathy
Liu	2019	44	F	RUQ pain, N/V, fever	MA, HyB		Cystic lesion in RHL w/ hemorrhage, PEL in A phase	cystic lesion w/ PEL		CBS w/o EL, HN	Resection	Diffuse bleeding	
Maruyama	2019	49	F	IF on CT	Nl	Mass w/ clear boundary, Nml flow on USD	PEL in A phase	T1HI, T2HI w/o SIPO enhancement	EGD	CBS		Lesions stable	Colon cancer, renal cell carcinoma
Angulo	2020	44	M	Elevated LFTs, jaundice	ALP 262, HyB, MA				EGD	FNH, CBS w/ fibrosis	Embol	Death	
Yoshida	2020	72	M	LLQ pain	Nl	HoL w/ irregular boundaries, no flow on USD	HEL	Halo-like lesion	FDG-PET, EGD	CBS			
Omata	2021	2	M	Fever, HS, ascites	LCT, MA, AST 237 ALT 227 LDH 453		SCH in RHL		Angiography		Hepatic artery embol, LDLT	HS, anemia, septic shock	Myotubular myopathy
Slouma	2021	55	M	Elevated LFTs	GGT 1014 ALP 278,					CBS	Infliximab	Nl LFTs	Psoriasis, topical steroids

*Literature review includes all published reports that were identified in English and able to be procured by our institution; Nl: normal; HM: hepatomegaly; HSM: hepatosplenomegaly; RHL: right hepatic lobe; HCC: hepatocellular carcinoma; FNH: Focal nodular hyperplasia; FNA: Fine Needle Aspiration; BMT: bone marrow transplant; LDLT: live donor liver transplant; HSCT: hematopoietic stem cell transplant; ITP: Immune thrombocytopenia purpura; OCP: oral contraceptive pills.

Table 2: Comprehensive literature review: Review articles*

Author	Year	Mean age	Cohort size	Sex ratio	Presenting symptoms	Appearance	Imaging	Treatment	Outcomes	Comorbidities
Breitschwerdt	2000									HIV/AIDS, *Bartonella* sp.
Morotti	2001		90							AIDS, CMV
Masaki	2002	61.1	8	M5, F3	Fever (2), liver enlargement (2), lymph node swelling (1), elevated ALP, LDH, CRP, ESR normal AST/ALT	Foam cells in portal tract, S100+, Birbeck granules, dilated lymph vessels	US	ABVD chemotherapy regimen (1) – vinblastine (1)	Mean survival 44 months, death from liver failure in 4 years	Hodgkin lymphoma (1) - diabetes insipidus (1), histiocytosis X (1)
Tsokos	2005					"Swiss cheese" parenchyma, irregular cavities w/o sinusoidal cells, fibrous tissue, phlebectatic w/ regular, spherical cavities			Spontaneous rupture	TB, hematologic malignancy, AIDS, post-transplant immunodeficiency, IVDA, chronic alcoholism, OCP/steroids
Iannaconne	2006				AS, IF, HM, ascites, portal HTN, cholestasis, HF, RUQ pain, lymphadenopathy (*B. henselae*), neuro sx (*B. quintana*)	Lesions involve entire liver or focal, CBS, bacillary peliosis stains w/ Warthin-Starry	US, Levovist-contrast US, CT, T2/T1 MRI	D/c offending drug or toxin, surgical resection, erythromycin (*Bartonella*)	Regression w/ d/c drug or infectious dx, hemorrhage, HF, portal HTN, liver rupture w/ HP, peliosis of spleen, lymph nodes and cutaneous lesions	Drugs (steroids, OCPs, tamoxifen, DES, 6MP, MTX), toxins (PVC, arsenic, thorium oxide), chronic wasting dx (TB, leprosy, malignancy, esp. HCC), AIDS, bacillary peliosis, post-renal or cardiac transplant
Modlinski	2006				IF	Sinusoidal dilation at peripheral zone of the hepatic lobule			Regression w/ d/c of drug, high mortality from internal hemorrhage, HF	Chronic Oral Anabolic steroids, biliary stasis, hepatomas
Gisbert	2007				Abnormal LFTs			Dose reduction or cessation	Regression of lesions	AZA, 6MP
Kim	2007		8				US (6), CT (8), MRI (3)			
Queiroz-Andrade	2009					Multiple CBS	T2 MRI, gadolinium MRI	Cessation of anemia treatment		Oxymetholone, iron transfusion

Table 2: *(Continued)*

Author	Year	Mean age	Cohort size	Sex ratio	Presenting symptoms	Appearance	Imaging	Treatment	Outcomes	Comorbidities
Franz	2011							Bacterial eradication w/ Abx		AIDS, *Bartonella henselae*
Rojas-Feria	2013									Inflammatory bowel dx, thiopurines (AZA, MP)
Srinivasa	2015		2		SHH		CT, Liver markers, coagulation studies, tumor markers		SHH	
Stine	2015				Chronic abnormal LFTs			Drug removal, immunosuppressive therapy		Drugs (amoxicillin-clavulanate, Bactrim, azithromycin)
Anstead	2016									Homelessness, HIV, *Bartonella quintana*
Elsayes	2017				AS, IF, HM, portal HTN, HF, ascites, RUQ pain, HR, HP	Fast-surge CEL, tubular, vessel-like, central target sign, no mass effect	US, contrast enhanced US, CT, MRI	D/c offending medication	Hemorrhagic parenchymal necrosis, death from hemorrhage	Idiopathic, steroids, OCPs, tamoxifen, MTX, arsenic, PVC, AIDS, TB, leprosy, malignancy, Bartonella, adenovirus, diabetes, pregnancy
Solimini	2017				Asymptomatic, HM			D/c anabolic steroids	Mortality from hemorrhage, HF	Aplastic anemia, Anabolic steroids, dietary supplements
Hui	2019				AS	Multiple lesions, no mass effect, lesions involve entire liver, or focal	Gadoxetic acid-enhanced MRI	Cessation of drug	Regression of lesions, HF, portal HTN, liver rupture w/ HP, rapidly fatal if untreated	Estrogen, progesterone, synthetic testosterone, OCPs
Demyashkin	2020									HCC, Kidney transplant, long-term immunosuppression
Shimizu	2020		2	M2, F0	SHH	Sinusoidal dilation w/ degenerative changes		Surgical resection, LDLT, embolization	Liver hemorrhage	X-linked myotubular myopathy
Siddiqi	2020			M 50% F 50%	IF, abnormal LFTs, RUQ pain, jaundice, portal HTN, infection		US, CT, MRI, angiography, biopsy	D/c offending medications, Abx therapy for bacillary infection	No dx progression, portal HTN, intraperitoneal hemorrhage	Medications (immunosuppression, tamoxifen, estrogens/androgens, OCPs), hematologic, transplant (renal), malignancy, autoimmune, TB, HIV, bacillus infection, X-linked myotubular myopathy, idiopathic
Luciani	2021	48	651	M9, F4	Fever		PCR sample of skin, liver or spleen	Doxycycline only (6), doxy + erythromycin (4), erythromycin only (1)		*Bartonella* sp., solid organ transplant, AIDS

*Literature review includes all published reports that were identified in English and able to be procured by our institution HM: hepatomegaly; HSM: hepatosplenomegaly; RHL: right hepatic lobe; HCC: hepatocellular carcinoma; FNH: Focal nodular hyperplasia; FNA: Fine Needle Aspiration; BMT: bone marrow transplant; LDLT: live donor liver transplant; HSCT: hematopoietic stem cell transplant; ITP: Immune thrombocytopenia purpura; OCP: oral contraceptive pills.

CONCLUSION

The dilatation of hepatic sinusoids in patients with PH poses a risk for morbidity and mortality due to spontaneous or surgically induced rupture and hemorrhage, necessitating early and accurate diagnosis. Unfortunately, most patients are asymptomatic, and, when diagnostic imaging such as sonography, CT, MRI, and angiography suspects PH, a biopsy is the only means by which to obtain a definitive pathologic diagnosis. However, as tissue procurement risks hemorrhage, and there is no consensus regarding standard treatment, patients should be evaluated initially for drug/toxicity-related PH for which regression may occur with removal of the offending agent. If there is no concern for malignancy, and for asymptomatic patients and those without indications of advanced liver disease, serial imaging and liver function testing may be appropriate management, with resection and embolization reserved for those with evidence of acute hemorrhage.

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Author Contributions

Alex Teshon – Acquisition of data, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Crystal Walker – Conception of the work, Acquisition of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

David P Schammel – Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Christine Marie-Gilligan Schammel – Design of the work, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

A Michael Devane – Conception of the work, Acquisition of data, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.