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Positron Emission Tomography/Computed Tomography and Magnetic Resonance Imaging Findings of Primary Leiomyosarcoma of the Liver: A Case Report and Literature Review

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Abstract

Background: Primary hepatic leiomyosarcoma (PHL) is an exceedingly rare malignant neoplasm, accounting for only 0.2% - 2% of all primary hepatic malignancies. Its non-specific clinical and imaging features often lead to misdiagnosis. **Aim:** This study aims to delineate the MRI and FDG PET/CT imaging findings in a patient with multiple primary hepatic leiomyosarcomas and to review the relevant English-language literature from 2000 to 2024, focusing on the MR and PET/CT characteristics of PHL. **Case Presentation:** We report the case of a 62-year-old male with no significant medical history who was incidentally found to have multiple intrahepatic lesions. Contrast-enhanced MRI revealed well-defined nodules showing T1 hypointensity, T2 hyperintensity, restricted diffusion, and progressive peripheral enhancement. FDG PET/CT demonstrated heterogeneous radiotracer uptake with SUVmax values ranging from 5.1 to 12.0. Histopathological examination of resected specimens confirmed the diagnosis of PHL. **Conclusion:** PHL, though typically presenting as a solitary mass, can manifest as multiple intrahepatic lesions. MRI is essential for lesion characterization, while PET/CT is valuable for staging and detecting extrahepatic disease. Awareness of its imaging spectrum is crucial for accurate diagnosis. Future integration of imaging omics and machine learning may further enhance diagnostic precision.

Keywords

Primary Hepatic Leiomyosarcoma, FDG PET/CT, MRI

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1. Introduction

Primary leiomyosarcoma of the liver is an exceptionally rare mesenchymal tumor, representing only 14% - 29% of all primary hepatic sarcomas, which themselves constitute less than 1% of all malignant liver tumors [1] [2]. Due to its rarity and non-specific clinical and radiological presentation, PHL is frequently misdiagnosed as other more common hepatic malignancies.

Imaging plays a pivotal role in the diagnostic workup. Magnetic resonance imaging (MRI) typically shows heterogeneous T2 hyperintensity and T1 hypointensity, with dynamic enhancement patterns that can vary. Positron emission tomography/computed tomography (PET/CT) is valuable for assessing metabolic activity, detecting distant metastases, and guiding biopsy. Previous literature has predominantly described PHL as a solitary large mass; however, multifocal presentation, as in our case, is exceedingly rare and warrants documentation.

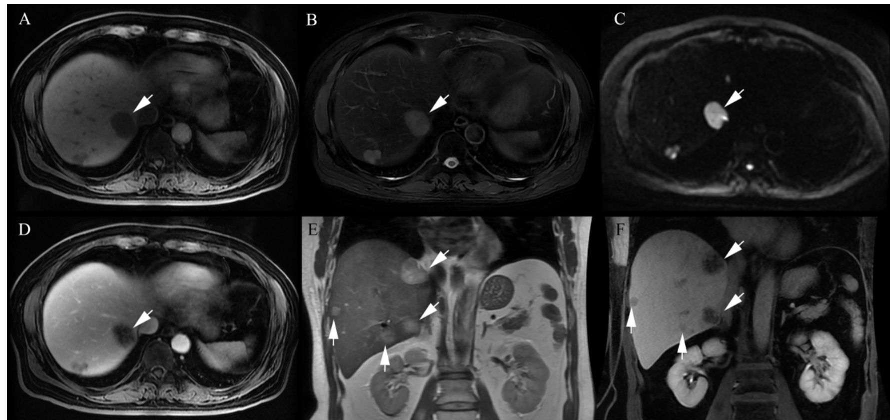
This study presents a detailed radiological description of a rare case of multifocal PHL, supplemented by a systematic review of the English literature from January 2000 to December 2024. Our objective is to enhance the radiological understanding of this rare entity, thereby facilitating accurate diagnosis and optimal patient management.

2. Case Description

A 62-year-old male patient was incidentally found to have multiple intrahepatic lesions during an ultrasound examination at a different hospital. He is asymptomatic and has no significant medical history, including liver diseases or alcohol abuse. His past medical and family histories are unremarkable. Physical examination revealed no abnormalities, and laboratory analysis showed a slightly elevated uric acid level. Other laboratory tests, including complete blood count, liver function tests, thyroid function tests, coagulation profile, and kidney function tests, were within normal limits. Tests for hepatitis B surface antigen, hepatitis C virus antibody, α-fetoprotein, and carbohydrate antigen 19-9 were negative.

Subsequently, contrast-enhanced magnetic resonance (MR) imaging of the abdomen was conducted to provide a more detailed characterization of the lesions (**Figure 1**). The abdominal magnetic resonance imaging (MRI) identified multiple intrahepatic nodules of varying sizes, with the largest measuring 36 mm × 30 mm × 33 mm. These lesions exhibited heterogeneous hypointensity on T1-weighted images and hyperintensity on both fat-suppressed T2-weighted and standard T2-weighted images, with necrosis or cystic changes observed in most lesions. The nodules were well-defined and lacked a discernible capsule. Additionally, most of the larger lesions displayed a hyperintense signal on diffusion-weighted imaging, and the corresponding apparent diffusion coefficient (ADC) map revealed decreased signal intensity in portions of the lesion walls. The enhancement scan revealed mild, heterogeneous rim enhancement during the arterial phase, followed by progressive and sustained enhancement characterized by a floral ring-like pattern at the periphery during the venous and portal phases. Within the lesion, short, strip-

like areas exhibited continuous and progressive enhancement. The majority of lesions demonstrated slightly reduced enhancement, while others maintained continuous enhancement on the 3-minute delayed scan. Furthermore, small blood vessels were observed encircling some of the lesions. No evident cancer thrombus was detected in the portal vein. A lymph node was identified in close proximity to the superior mesenteric artery.



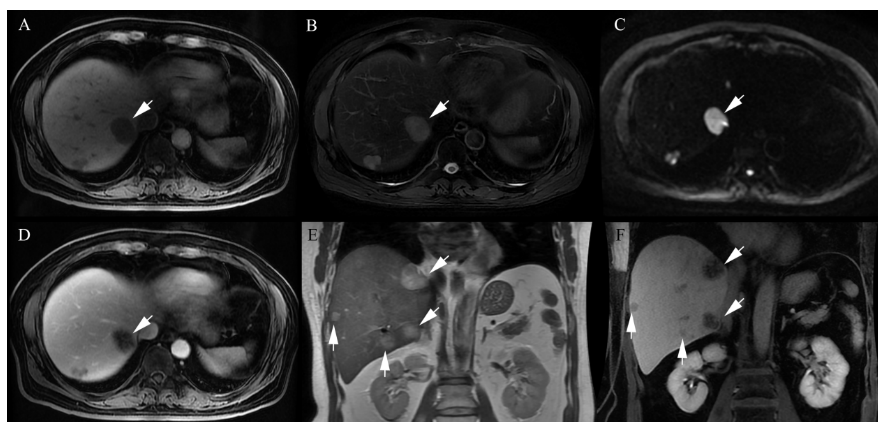
MR imaging reveals multiple intrahepatic nodules of varying dimensions. The largest lesion is well-defined and shows slightly heterogeneous hypointensity on T1-weighted imaging (A, arrow) and heterogeneously hyperintense on T2-weighted imaging (B, arrow).On diffusion-WI, the lesion displays hyperintensity (C, arrow).In the late arterial phase of dynamic contrast-enhanced imaging, the lesion shows heterogeneous rim enhancement (D, arrow). Coronary T2WI showed multiple intrahepatic lesions (E, arrows), and they all show marginal enhancement during the arterial phase (F, arrows).

Figure 1. MRI findings of primary liver leiomyosarcoma.

The patient was referred for an ^{18}F-FDG positron emission tomography-computed tomography (PET-CT) whole body scanner to determine the origin of the lesions and identify other metastatic sites for accurate disease staging prior to surgery (**Figure 2**). The PET-CT revealed multiple lesions with heterogeneous FDG uptake on the maximum intensity projection image, exhibiting SUVmax values ranging from 5.1 to 12.0. These lesions corresponded to the tumors identified by MRI. The largest lesion, located in segment IV, demonstrated significant activity with peripheral radioconcentration characterized by an SUVmax of 7.0 and central radioconcentration with an SUVmax of 5.1. The lesion with the highest radioactive uptake was located in segment VI, exhibiting an SUVmax of 10.5. Additionally, an enlarged lymph node adjacent to the superior mesenteric artery, measuring approximately 24 mm × 21 mm, showed radioconcentration with an SUVmax of 6.8. The presence of multiple intrahepatic lesions and an enlarged lymph node adjacent to the superior mesenteric artery suggests the existence of multiple primary malignant tumors. Furthermore, there was increased radioactivity distribution in the soft tissues surrounding the left knee and ankle joints.

Subsequently, the patient underwent a partial hepatectomy involving segments S4 and S6. Pathological examination of the excised liver tissue confirmed a diagnosis

of primary hepatic leiomyosarcoma. Notably, the diagnosis of primary hepatic leiomyosarcoma was based on the histological features of the liver lesions, which were characteristic of a primary smooth muscle tumor, and the absence of any identifiable primary extrahepatic leiomyosarcoma upon comprehensive clinical and radiological evaluation, including the whole-body PET/CT. After a definitive diagnosis was established, the patient commenced comprehensive anticancer treatment.



Three-dimensional maximum intensity projection (A) showed multiple lesions demonstrating heterogeneous FDG uptake(arrows). The transaxial images (B) and (C) of the ^{18}F-FDG PET-CT demonstrated the lesions in the largest size and in the highest radioactive value (arrows). There was also increased radioactivity distribution in the left knee joint and ankle joints, which is indicative of degenerative processes.

Figure 2. PET/CT findings of primary liver leiomyosarcoma.

3. Discussion

Primary hepatic leiomyosarcoma is an exceptionally rare malignancy worldwide, constituting only 14% - 29% of all primary hepatic sarcomas, which themselves represent less than 1% of all malignant hepatic tumors [2]. Other types of primary hepatic sarcomas include angiosarcoma, fibrosarcoma, liposarcoma, embryonal sarcoma, malignant fibrous histiocytoma, carcinosarcoma, and epithelioid hemangioendothelioma [3]. It is hypothesized that primary hepatic leiomyosarcoma originates from smooth muscle cells located within intrahepatic vessels, bile ducts, or ligaments [1]. The clinicopathological characteristics, imaging features, treatment modalities, and prognosis of this malignancy remain poorly understood.

3.1. Clinical Manifestations of PHLS

Primary hepatic lymphoma (PHL) exhibits a broad age distribution, with the youngest reported patient being 5 months old and the oldest 86 years old. While PHL predominantly affects older adults, with a mean age of onset at 52.4 years [2], it can also occasionally manifest in pediatric populations [4]-[6]. The prevalence of PHL is slightly higher among female patients.

The clinical manifestations of primary hepatic lymphoma (PHL) are typically non-specific, with tumors generally remaining asymptomatic until they reach a

significant size. Common symptoms include abdominal pain, abdominal distension, nausea, jaundice, weight loss, and vomiting. In some cases, patients may also present with fever, potentially associated with tumor hemorrhage or infection [7] [8]. Physical examination often reveals hepatomegaly and a palpable mass [9]. Notably, tumor marker levels such as alpha-fetoprotein (AFP), carcinoembryonic antigen (CEA), and carbohydrate antigen 19-9 (CA19-9) usually remain within normal limits [10].

The precise pathogenetic mechanisms underlying the condition remain unidentified. Certain cases of primary hepatic lymphoma (PHL) have been documented in close association with immunocompromised states, such as AIDS [11] [12], the post-renal transplant period [13] [14], radiochemotherapy for Hodgkin's lymphoma [15], and liver-related diseases, including chronic hepatitis C [16] and hepatitis B [17]. This association is hypothesized to result from the unrestrained effects of the Epstein-Barr virus (EBV) on smooth muscle proliferation [11]. Nevertheless, a significant proportion of patients diagnosed with primary hepatic lymphoma were immunocompetent, with approximately 77.4% exhibiting no discernible predisposing factors [2] [12]. Consequently, the etiology of primary hepatic lymphoma syndrome (PHLS) remains ambiguous and warrants further investigation.

The current patient, an elderly male, exhibited no abdominal discomfort and demonstrated normal levels of tumor markers. His medical history is unremarkable, with no previous liver diseases or immunodeficiency disorders. Laboratory tests were largely unremarkable, except for a mild elevation in uric acid levels.

Due to the non-specific nature of the clinical manifestations and laboratory findings, primary hepatic leiomyosarcoma (PHL) is frequently misdiagnosed [10] [18]. Accurate preoperative diagnosis of hepatic leiomyosarcoma, including identification of primary sites and potential metastasis, is crucial [12] [19]. Precise diagnostic protocols are essential for developing effective treatment strategies, particularly in the context of neoadjuvant therapy and complex surgical planning with extended margins, to address the aggressive nature of PHL. Imaging modalities are integral in this context, with magnetic resonance imaging (MRI) being essential for the diagnosis of liver lesions and positron emission tomography/computed tomography (PET/CT) playing a critical role in the detection of primary lesions and their metastases. In this study, we present a case of primary hepatic lymphoma (PHL) and conduct a thorough review of the existing literature to raise awareness about this rare malignant tumor. Our aim is to advance the radiological understanding of PHL, thereby optimizing patient management and improving clinical outcomes.

3.2. Literature Review

Case reports and case series pertaining to PHLS, published in English from January 1, 2000, to December 31, 2024, were sourced from PubMed and Google Scholar. After a rigorous screening process, 23 publications comprising 24 cases that described the MR imaging and PET/CT characteristics of PHLS were selected for

inclusion in this study. The imaging findings from these cases, along with the current case, are summarized in **Table 1**.

Table 1. Imaging findings of PHLS from the literature and the present patient.

References	Case	Size (mm)	Location	Margin	MR							
					T1WI	T2WI	Hemorrhage or/and Necrosis	Envelope	DWI	Enhancement	PET/CT	Metastasis
[7]	1	97	Right lobe		Heterogeneous hypointensity	Heterogeneous hyperintensity	Hemorrhage	NA	Restricted diffusion in the wall of the lesion	AP: the edges and septations significantly enhanced VP, DP: fused and filled toward the center	Increased uptake	(-)
[27]	2	25, 40	Segments IV and VII	Well-defined	NA	NA	NA	NA	NA	NA	Mild hypermetabolic	Bone metastasis
[21]	3	110	Left lobe	Well-defined	Slightly heterogeneous hypointensity	Heterogeneous hyperintense	NA	NA	Restricted diffusion	AP&VP: no evident enhancement DP (3-min): marked enhancement	NA	Intrahepatic and right adrenal metastases
[2]	4	68	Left lobe	Well-defined	Hypointensity with internal hemorrhage	Heterogeneous hyperintensity with a hypointense area	Hemorrhage and necrosis	NA	Restricted diffusion	AP: heterogeneous enhancement PVP&DP: gradual enhancement	NA	
[10]	5	124	Right lobe	Well-defined	NA	Hyper-	NA	NA	NA	NA	NA	NA
[32]	6	120	Lateral segment	Well-defined	Homogenous hypointense	Hyperintense	NA	NA	NA	AP: heterogeneously enhanced DP: weakly enhanced during the late phase	Higher ^{18}F-FDG uptake	Lungs and intrahepatic metastasis
[9]	7	198	Right lobe	NA	Heterogeneous hypo-	Slight hyperintensity with central heterogeneous hyper- & hypo-intensity	Necrosis	NA	Restricted diffusion	AP-PVP: peripheral gradual enhancement	Sizeable focus of intense accumulation of radiolabelled glucose	(-)
[33]	8	40	Caudate lobe	NA	Hypo-	Heterogeneous hyper-	Hemorrhage and necrosis	(-)	Heterogeneous high & low	Gradual mild enhancement	NA	Intrahepatic metastases
[20]	9	57	Left lobe	Well-defined	Heterogeneous hypo-	Hyper-	NA	(-)	NA	Discretely hypervascular	NA	None
	10	290	Right lobe	NA	Hypo-	Hyper-	NA	NA	NA	AP-PVP: peripheral gradual enhancement	NA	Lungs metastases
[28]	11	113	Segment IV, V, and VI	Well-defined	Hypo-	Mild hyper-	NA	NA	Mild diffusion restriction	Gradual enhancement	Low-Fluorodeoxyglucose (FDG) avid	NA
[34]	12	130	Left lobe segments III and IVB	Irregular margin	Heterogeneous hypo-	Heterogeneous hyper-	NA	NA	N/A	N/A	NA	Multiple abdominal metastases
[4]	13	70	Hepatic hilum	Well-defined	Hypo-	Heterogeneous hyper-	NA	NA	Restricted diffusion	N/A	Focally increased activity	(-)Iida, T. 2017
[8]	14	100	Left lobe	NA	Slightly hypo-	Slightly hyper-	NA		NA	NA	Higher ^{18}F-FDG uptake	N/A
[22]	15	91	Caudate lobe and the left lobe	Well-defined	Slightly heterogeneous hypo-	Heterogeneous hyper-	NA	(-)	Hyper-	AP&VP: no evident enhancement DP (5-min): marked enhancement	Higher ^{18}F-FDG uptake	Intrahepatic metastases
[11]	16	90	Left lobe	Well-defined	Hypo-	Hyper-	Necrosis	N/A	NA	Irregular peripheral enhancement	N/A	N/A

Continued

[35]	17	137	Right lobe	Well-defined	Centrally increased signal consistent with internal hemorrhage	Hypointense rim compatible	Hemorrhage	Capsule or pseudoca psule	NA	Peripheral enhancement	N/A	N/A
[36]	18	42	Left lobe	Well-defined	Hypo-	Hyper-	NA	N/A	Hyper-	Delayed washout or gradual enhancement	N/A	Intrahepatic metastases
[37]	19	174	Right and left lobes	N/A	Heterogeneous hypo-	Heterogeneous hyper-	Necrosis or hemorrhage	N/A	NA	Peripheral enhancement	N/A	N/A
[38]	20	230	Right lobe	N/A	Hypo-	Hyper-	NA		NA	Peripheral enhancement		
[39]	21	140	Left lobe	Well-defined	Hypo-	Hyper-	NA		NA	Peripheral enhancement		
[40]	22	350	Left lobe	NA	Hypo-	Peripheral isointensity with central hyperintensity	NA		NA	Peripheral enhancement		
[14]	23	50	Right lobe	Well-defined	Hypo-	Hyper-	NA		NA	NA		
[16]	24	140	Right lobe	Well-defined	Hypointensity with internal hemorrhage	Heterogeneous hyperintensity with a hypointense area	NA		Restricted diffusion	AP: heterogeneous enhancement PVP&DP: gradual enhancement		
Ours	25	36	Right and left lobes	Well-defined	Hypo-	Hyper-	Necrosis	(-)	Restricted diffusion	AP-PVP: peripheral gradual enhancement	Higher ^{18}F-FDG uptake	Lymph node metastasis

3.3. Imaging Findings of PHLS

The non-specific imaging characteristics of primary hepatic leiomyosarcoma (PHL) pose significant challenges for accurate preoperative diagnosis. Traditional ultrasonography (US) generally identifies a hypoechoic mass, while computed tomography (CT) scans predominantly reveal well-defined hypo- or iso-dense masses, often containing heterogeneous regions suggestive of necrosis or hemorrhage [2].

Magnetic Resonance Imaging (MRI) typically reveals heterogeneous hyperintensity on T2-weighted imaging (T2WI) and hypointensity on T1-weighted imaging (T1WI). This imaging modality may also display internal heterogeneity, indicative of intratumoral hemorrhage or necrosis [9]. Diffusion-weighted imaging was conducted in 10 out of 24 patients, showing varying degrees of restricted diffusion in all cases. Notably, one case exhibited restricted diffusion with wall involvement [7]. Most tumors were well demarcated, lacking a distinct capsule. A review of the literature indicates that a significant proportion of these cases demonstrate peripheral, heterogeneous enhancement during the arterial phase, with persistent or gradual enhancement noted in the portal venous or delayed phases [7] [9] [20]. In certain instances, there was an absence of enhancement during the arterial and venous phases, with only mild enhancement observed in the delayed phase [21] [22]. The MRI features in our case align with the predominant literature findings, particularly the progressive delayed enhancement, which may be a suggestive feature of PHL. A notable aspect of our case is the multifocal presentation, which contrasts with the more common solitary large mass. This

multifocality could imply a different growth pattern or potentially a more aggressive tumor biology, though more cases are needed to establish any definitive clinicopathological correlation. Moreover, the lesions in this case uniformly exhibited similar MR imaging characteristics.

As a non-invasive diagnostic technique, PET/CT is utilized for the detection of primary tumors and metastases, as well as for distinguishing between benign and malignant lesions with high sensitivity. Additionally, it is employed for staging and guiding biopsy procedures [23]. Previous studies have reported FDG PET/CT findings in cases of leiomyosarcoma affecting the liver and other rare organs [24] [25] [26]. In the literature, the majority of primary hepatic leiomyosarcoma (PHL) cases exhibited high FDG uptake, with only one case demonstrating mild hypermetabolism comparable to the metabolic activity of liver parenchyma [27]. Another case showed low FDG uptake [28]. Cheon [27] suggested that the value of metabolic parameters such as SUVmax in the 18F-FDG PET-CT may be linked to the level of differentiation or necrosis and that it might also help diagnose and stage leiomyosarcoma. The substrates associated with varying FDG uptake remain ambiguous. In alignment with the majority of previous studies, the current case, characterized by multiple intrahepatic lesions, demonstrated elevated radiotracer uptake, with the SUVmax values at the periphery of most lesions exceeding those at the center. An increased distribution of radioactivity was observed in the soft tissues surrounding the left knee and ankle joints, with an SUVmax of 15.7, potentially indicative of bursitis or degenerative processes. Furthermore, no lesions were identified in other organs, allowing for the exclusion of hepatic metastases. Nonetheless, additional data collection is necessary for a more comprehensive evaluation.

Our findings suggest that primary hepatic leiomyosarcoma may present as multiple intrahepatic lesions. The diagnosis of primary hepatocellular sarcoma is confirmed through the histopathological analysis of surgical specimens or autopsy samples. Postoperative surveillance of prognosis and disease progression requires the utilization of MRI or PET-CT imaging modalities.

4. Conclusion

We present a rare case involving multiple intrahepatic leiomyosarcomas. Primary hepatic leiomyosarcoma (PHL) should be considered in the differential diagnosis when encountering heterogeneous lesions with a rich blood supply and persistent delayed enhancement of the solid component. It is important to note that while PHL typically manifests as a solitary large lesion, it can also present as multiple intrahepatic lesions. Magnetic Resonance Imaging (MRI) is crucial for preoperative or pre-treatment assessment, and Positron Emission Tomography/Computed Tomography (PET/CT) is essential for identifying the primary lesion. Currently, the advent of imaging omics has significantly enhanced diagnostic capabilities for various diseases. Looking ahead, radiomics and machine learning present promising solutions for the diagnostic challenges of PHL. By extracting subvisual char-

acteristics from MRI and PET/CT images, these technologies can build intelligent models to accurately differentiate PHL from other hepatic tumors, including hepatocellular carcinoma (HCC), intrahepatic cholangiocarcinoma (ICC), and metastases. Furthermore, they hold potential for non-invasively predicting tumor behavior and treatment response, paving the way for precision medicine and personalized management of this rare disease [29]-[31].

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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