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Case Report

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A Case of Pedunculated Focal Nodular Hyperplasia Treated by Laparoscopic Hepatectomy

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Abstract

Focal nodular hyperplasia is the second most common benign liver tumor after liver hemangioma. Focal nodular hyperplasia has essentially no potential for malignant transformation, but hemorrhage may occur in rare cases. We describe herein our experience performing laparoscopic resection in a patient with pedunculated focal nodular hyperplasia. We also discuss the relevant literature on pedunculated focal nodular hyperplasia, given the rarity of this type of tumor.

Keywords: Exophytic; FNH; Laparoscopic Resection; Projected; Pedunculated

Introduction

Focal nodular hyperplasia (FNH) has traditionally been classified as a type of benign hepatocellular nodule, and is defined as a hyperplastic lesion caused by vascular anomaly and characterized by normal background liver, a grossly visible central scar, and histologically evident anomalous blood vessels and portal tract [1]. FNH poses very little risk of malignant transformation or hemorrhage, but occasional reports have described actual cases of FNH in which differentiation from hepatocellular carcinoma (HCC) prior to surgery was difficult [2,3].

The present study describes our experience with a patient who presented with typical FNH characteristics in diagnostic imaging prior to surgery, underwent laparoscopic hepatectomy, and was ultimately diagnosed with FNH on pathological examination.

Case Report

The patient was a 39-year-old man with no previous medical history of note other than childhood asthma. He underwent medical exam at a local clinic and abdominal ultrasonography performed revealed a 35-mm mass lesion in the liver. So he was referred to the gastroenterology department at our hospital for detailed examination and treatment. He had consumed 28 g/day of alcohol (700 mL beer/day) between 20 and 37 years old, and had been smoking 5 cigarettes a day for the past 15 years. He had no history of medication use. Hematology did not reveal anything of note (Table 1). Abdominal ultrasonography showed a 39 × 30 × 37-mm hypoechoic nodular tumor that appeared to extend from the caudal aspect of liver segment 6. Blood flow was observed within the tumor that appeared isoechoic to background liver (Figure 1). Dynamic CT of the liver showed a 32-mm tumor with suspected hepatic origin near the hepatic flexure of the colon that was densely stained in the early phase and showed iso-density in the late phase. The vein draining the tumor was connected to the hepatic vein (Figure 2). Moreover, dynamic MRI (Gd-EOB-DTPA) of the liver revealed a tumor which showed iso-density in T1W1 inphase, T2W1 and hepatobiliary phase and which showed similar finding to when dynamic CT was done and had a central scar leading to a suspected diagnosis of FNH (Figure 3). Differential diagnoses such as Angiomyolipoma (AML), hemangioma, Gastrointestinal Stromal Tumor (GIST) and Neuroendocrine Tumor (NET) of enteric origin were also considered.

WBC	5900	/μl
Hb	14.8	g/dl
PLT	270,000	/μl
PT	105	%
TBil	0.56	mg/dl
AST	17	U/l
ALT	11	U/l
TP	7.6	g/dl
Alb	4.7	g/dl
BUN	13	mg/dl
Cr	0.85	mg/dl
Na	141	mmol/l
K	4.7	mmol/l
Cl	104	mmol/l
AFP	3	ng/ml
PIVKA-II	12	mAU/ml
CEA	1.4	ng/ml
CA19-9	4	U/ml
NSE	9.1	ng/ml
HBs-Ag	-	
HBs-Ab	-	
HBc-Ab	-	
HCV-Ab	-	

Table 1: Laboratory data at first visit.

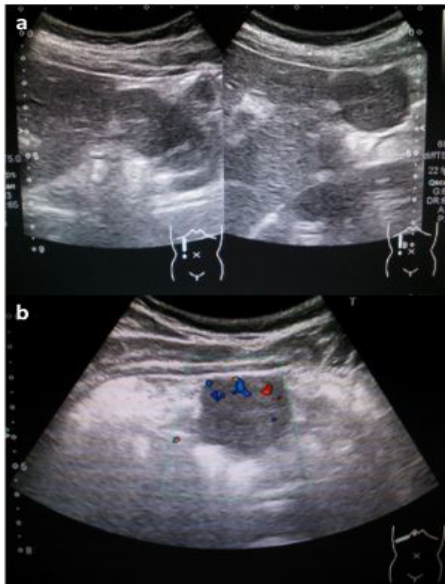

Figure 1: Ultrasound (a) B-mode and (b) Color Doppler A 39 × 30 × 37-mm hypoechoic nodular tumor extending from the caudal aspect of liver segment S6 (a). Blood flow is apparent within the tumor (b).

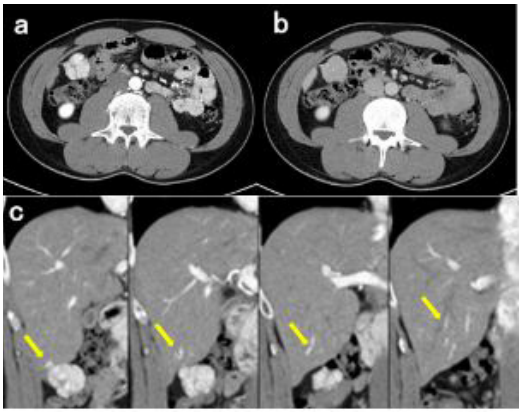

Figure 2: Dynamic CT A 32-mm tumor of suspected hepatic origin near the hepatic flexure shows dense staining in the early phase (a), and showed iso density in the late phase (b). Drainage vein of the tumor connected to right hepatic vein in early phase (arrow) (c).

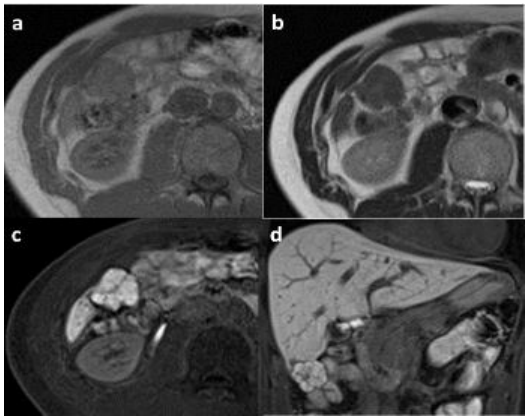

Figure 3: Gd-EOB-DTPA-enhanced MRI, (a) T1 W1 inphase, (b) T2 W1 Dynamic MRI of the liver revealed a tumor which showed iso-density in T1W1 inphase (a), T2W1 (b) and hepatobiliary phase (c) (d) and had a central scar.

Given the presence of a central scar, the fact that the hepatic vein was the drainage vein, the presence of dense early-phase staining, and the isointensity of the tumor to the liver in the hepatocellular phase on MRI, FNH was considered as the most likely diagnosis. However, due to the peculiar pedunculated morphology and risk of hemorrhage of the tumor, as well as the fact that malignant disease could not be completely ruled out, the decision was made to perform laparoscopic partial hepatectomy. The tumor was protruding from liver segment S6 and was 3 cm in diameter. Liver parenchymal transection was performed using a harmonic scalpel. Once transection had been made deep into the parenchyma, the vasculature and drainage vein entering the tumor became visible. After ligating and then cutting the vasculature and drainage vein using the harmonic scalpel, the tumor was resected. Macroscopically, the resected tumor specimen was a yellowish-brown, multinodular mass lesion with partial white fibrosis, with a border clear-

ly distinguishable from the background liver. Histologically, the mass was unevenly demarcated by fibrous septa, and misaligned hepatic cords were also visible. Sinusoids were slightly dilated and hepatocytes at this site tended to be mildly enlarged, with some exhibiting enlarged nuclei. However, the nuclear-cytoplasmic ratio was not elevated, and no cellular atypia suggestive of tumor was evident. Lymphocytic infiltration, bile ductular hyperplasia, and abnormal muscle blood vessels were observed in the fibrous stroma, and FNH was diagnosed (Figure 4).

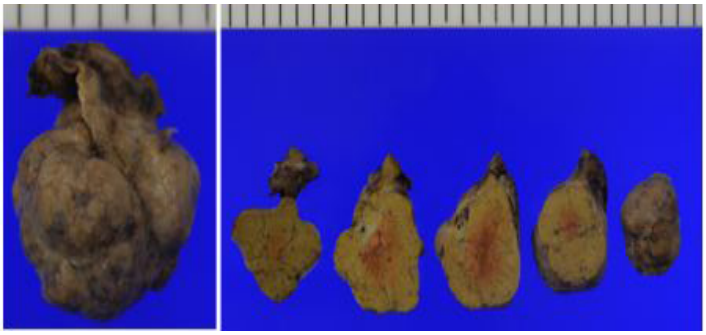

Figure 4: Macroscopic pathological findings Macroscopically, the resected tumor specimen is a yellowish-brown, multinodular mass lesion with partial white fibrosis, and a border that is clearly distinguishable from background liver.

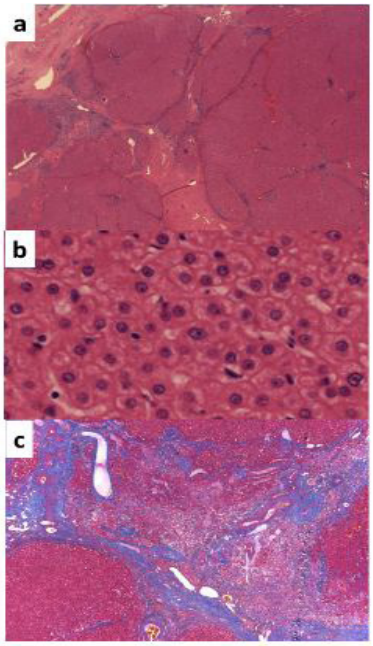

Figure 5: Microscopic pathological findings. The mass is unevenly demarcated by fibrous septa, misaligned hepatic cords are visible, and sinusoids are slightly dilated in Hematoxylin and eosin stain (a). Hepatocytes at this site tend to be mildly enlarged, with some exhibiting enlarged nuclei., but no cellular atypia suggestive of tumor is evident (b). Bile ductular hyperplasia, and abnormal muscle blood vessels are observed in the fibrous stroma in Azan stain (c).

Discussion

FNH is the second most common benign liver lesion after hemangioma [4,5]. This pathology mainly affects women in their thirties and forties [6], with a reported sex ratio between men and women of between 1:8 and 1:12 [4,7,8]. The pathogenic mechanisms underlying FNH are not yet completely understood. One theory is that circulatory failure brought on by a congenital arterial anomaly triggers hepatocellular hyperplasia, and this putative mechanism is considered responsible for the formation of hyperplastic nodules accompanied by the central scar [4] Most cases of FNH are asymptomatic, and rupture or hemorrhage are reportedly very rare [4,9]. However, scattered reports have described FNH rupture [3,10], and Li et al. recommended surgical resection of FNH ≥5 cm in diameter [3]. On the other hand, a separate study described a watchful waiting approach to an FNH 9 cm in diameter [11], suggesting the need for further debate.

Moreover, although FNH typically does not involve malignant transformation [4,8,9] one study reported two cases of malignant transformation among ≥800 cases of FNH [12] Generally, surgical treatment is often considered for the purpose of a definitive diagnosis or when symptoms are present [13]. On ultrasonography, FNH presents with internal echo patterns varying from low to high echogenicity. On CT, FNH appears densely stained during the early phase and varies from low density to slightly high density in the late phase. On MRI, FNH may appear hypo- to iso-intense on T1-weighted imaging and hyper-intense on T2-weighted imaging, but is not associated with any characteristic findings. The central scar appears as either a hyperechoic or hypoechoic region on ultrasonography, and as a low-density area in the arterial-dominant phase and a slightly contrast-enhanced area in the equilibrium phase of CT, but the visualization rate is not very high, at approximately 50-60%. The visualization rate of the central scar on MRI is reportedly about 80% [6]. A combination of MRI and other imaging modalities can allow a ≥90% sensitivity and specificity for FNH diagnosis [13]. However, a few case reports published in Japan have described the difficulties involved in preoperative diagnosis [2,3]. In addition, the rate of correct preoperative diagnosis has been reported as 59.3% in patients undergoing FNH resection [14].

Morphologically, the patient described in the present study represented a very rare case in that the FNH was protruding from the liver. A PubMed search using the terms “Projected FNH,” “Pedunculated FNH,” and “Exophytic FNH” revealed only five cases reported in English, indicating this as a very rare manifestation (Table 2) [15-19]. Conversely, a study reviewing the findings of 78 patients undergoing FNH surgery found that exophytic growth occurred in 40 lesions [20], although the extent to which these lesions protruded from the liver was unclear. In a study by Bieze et al. that included 59 FNH patients, conservative treatment was provided to 43 of these patients (73%), while 11 patients underwent resection

via laparotomy and 5 underwent laparoscopic resection. The authors found that postoperative length of stay was significantly shorter in patients who underwent laparoscopic surgery than in those who underwent laparotomy [21]. Meanwhile, in a study by Descottes, et al. 12% of FNH patients scheduled to undergo laparoscopic hepatectomy were switched to laparotomy for reasons such as serious bleeding and unfavorable lesion site [13,22].

	Year/Author	Age/ Sex	Size/form	Location	Symptom/ complication		Treatment
case 1	2001 Till R. Bader et al.	Unknown	4.5 cm exophytic	S5	Upper abdominal pain	Unknown	Surgery
case 2	2004 Valerie Byrnes	30/ F	①16×18 cm exophytic ②3.5×3 cm pedunculated	①S4 ②S3	Right upper quadrant pain	FNH[1)]	Laparotomy
case 3	2008 Nabil Wasif et al.	48/ F	3.2×2.4×2.4 cm pedunculated	S4	Intermittent right upper-quadrant pain	Uncertain	Laparoscopic resection
case 4	2012 Tadashi Terada	26/ F	4×4×5 cm projected	Left lobe	No symptom	GIST[2)]	Laparoscopic resection
case 5	2015 Radu Badea, et al.	29/ F	8×5 cm pedunclated	S5	Pain in the right hypochondrium and the umbilical area	Benign focal hyperplasia	Operation performed by laparoscopically
case 6	2014 our case	39/ M	39×30×37 mm pedunculated	S6	No symptom /no complication	FNH	Laparoscopic resection

[1)]FNH: Focal Nodule Hyperplasia; [2)]GIST: Gastrointestinal Stromal Tumor.

Table 2: Characteristics of projected, pedunculated or exophytic FNH in the literature.

In the present patient, we initially surmised that the mass represented a typical case of FNH based on the preoperative imaging findings of the presence of a solitary peripheral lesion accompanied by a central scar and lacking portal blood flow, as well as the lack of accompanying portal hypertension and the absence of background liver cirrhosis. However, we later opted for surgical resection given the very rare morphology of the FNH in terms of protrusion from the liver surface, the inability to completely exclude the malignancies GIST and NET on dynamic MRI, and the potential risk of hemorrhage, as well as reports in the literature of malignant transformation (although rare) even if the lesion did represent FNH. As laparoscopic resection is less invasive, and because the morphological characteristic of pedunculated growth was considered to facilitate successful removal, we performed laparoscopic resection. Hematoxylin and eosin staining of histopathological sections of the resected specimen then led to a diagnosis of FNH.

Resection should be considered when FNH proves difficult to diagnose or differentiate from malignant disease, or when the patient is symptomatic or carries a risk of events such as hemorrhage. In these scenarios, laparoscopic resection has the merits of being minimally invasive and enabling definitive diagnosis to be made using the resected pathological specimen.

The present study describes our experience performing laparoscopic resection of an FNH that was suspected to be a classic manifestation of this tumor based on diagnostic imaging prior to surgery.

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