Pedunculated hepatic focal nodular hyperplasia: A case report and review of the literature

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Abstract
Focal nodular hyperplasia (FNH) is a common asymptomatic benign hepatic tumor encountered in middle-aged women. However, pedunculated FNH is exceedingly rare and more frequently associated with complications. That is why surgical management is mandatory in this form.

KEYWORDS
focal nodular hyperplasia, liver resection, pedunculated liver tumor

1 | INTRODUCTION

We describe a case of pedunculated hepatic focal nodular hyperplasia (FNH) in a 38-year-old woman, incidentally discovered at the surgery when repairing an epigastric hernia. The tumor was attached to segment 3 of the liver by a stalk. Complete surgical resection was performed. The pathological findings complied with the diagnosis of FNH.

FNH is a benign hepatic neoplasm accounting for 3%-5% of all primary hepatic tumors,1 second in frequency after hemangioma.2 It is asymptomatic and commonly encountered in women during the third to fifth decades of life.3

FNH is located within the liver parenchyma in 80% of the cases (typical forms).4 FNH presenting with an exophytic growth is called pedunculated, and they represent the rarest atypical form5 with less than ten cases reported in the literature. This form is associated with a challenging diagnosis and requires surgical treatment.6

We herein present the case of a patient with an incidental finding of a pedunculated hepatic mass which turned out to be FNH. We also discuss the relevant literature on pedunculated FNH, given the rarity of this type of tumor.

2 | CASE REPORT

A 38-year-old woman without a significant medical history was admitted to the general surgery department for the management of a recurrent epigastric hernia. Clinical examination showed a noncomplicated epigastric hernia measuring about 6 cm in diameter. It was painless and utterly reducible with an expansile cough impulse. Blood investigations were within normal ranges. An abdominal ultrasound was done as part of the preoperative check-up and did not show any abnormal findings. Thus, an elective surgical repair of the hernia was scheduled, and a transabdominal mesh was planned. A midline epigastric incision was performed. The hernial sac of 8 cm long was identified and opened after circumferential dissection. It contains omentum of good vitality which was easily reduced into the abdominal cavity revealing a 6 cm fascial defect. Intraoperatively, a lobulated 3 cm exophytic mass arising from segment III of the liver was incidentally identified (Figure 1). This liver mass was perhaps missed by the radiologist. The lesion was attached to the liver by a thin stalk. As the patient had been on oral oestroprogestative contraceptives for three years,
the diagnosis of a pedunculated FNH was suspected. Since the diagnosis of hepatocellular adenoma or carcinoma cannot be completely ruled out, the decision was to resect the tumoral mass. A suture repair of the parietal defect was performed, and the placement of a transabdominal mesh was postponed.

The macroscopic examination of the resected specimen revealed a nodular mass measuring 4.5 × 3.5 × 3 cm with a central fibrous scar (Figure 2). Histologically, this central scar responds to a fibrovascular focus surrounded by multiple radial projections that incompletely circumscribe hepatic lobules. These fibrous septa are traversed by a fairly dense lymphocytic inflammatory infiltrate and include a hyperplastic cholangiolar proliferation at the periphery of the lobules (Figure 3).

This pathological finding was compatible with the diagnosis of a pedunculated FNH of the liver. The postoperative course was uneventful, and the patient was discharged on the 5th postoperative day. The use of oestroprogestative contraceptives was interrupted postoperatively, and a uterine device was implemented. No recurrence of the epigastric hernia or the liver mass was detected at the 2-year follow-up.

3 | DISCUSSION

FNH is the second most common benign hepatic tumor after hemangioma.2 It arises more often in the right lobe of the liver.7 The typical form (subcapsular) of FNH is most of the time asymptomatic and generally incidentally discovered in middle-aged woman.3 Pedunculated FNH is an atypical form of FNH located in the extrahepatic region and connected to the liver by a thin stalk. FNH accounts for up to 8.8% of all primary hepatic tumors; among them, the pedunculated form represents only 3% according to the results of the large study of Schild et al.5

A PubMed search using the terms “Projected FNH,” “Pedunculated FNH,” and “Exophytic FNH” revealed only a few cases reported in the English literature (Table 1).

Clinically, pedunculated FNH is more likely to be symptomatic due to the possible compression of the surrounding vessels by a large size tumor, gastric outlet obstruction by pedunculated tumors of segments 2, 3, 4, and 5, and hemorrhagic complications due to the rupture of the feeding vessel of the thin stalk connecting the tumor (FNH) to the liver.8

Moreover, a particular complication of pedunculated FNH should be kept in mind which is the torsion of the pedicule leading to infraction of the tumor if not early diagnosed.8

The diagnosis of FNH is frequently guided by radiographic investigations combining ultrasound, CT scan, and MRI. The latter is characterized by its highest sensitivity and specificity.9 The typical features of FNH seen on MRI include a mass that is iso- to hypointense on T1-weighted images, is iso- to hyperintense on T2-weighted images, and has avid enhancement on arterial phase imaging and isointensity on portal venous phase imaging. The central scar, when present, classically appears hypointense on T1-weighted imaging, hyperintense on T2-weighted imaging, and hyperintense on equilibrium-phase imaging.10

Except for the location, pedunculated FNH has similar imaging features to the typical form of FNH. However, their preoperative diagnosis is challenging as the pedicule is not easily identified radiologically.11

In the present patient, the diagnosis was made intraoperatively, and the tumor was incidentally discovered when
repairing an epigastric hernia. The radiology workup was limited to conventional abdominal ultrasonography that did not objectify the liver mass.

This fact can be explained by the lack of experience of the radiology resident who did perform the ultrasound.

Conservative management which consists mainly in the cessation of oral contraceptive pills is recommended for the typical forms of FNH. Surgical resection is indicated only in symptomatic, complicated, and atypical forms.6

For the rare forms of pedunculated FNH, surgery is required to prevent the complications mentioned above and to establish the final diagnosis using the resected pathological specimen. Currently, the laparoscopic approach of these tumors is considered to be a feasible and safe technique.12

| TABLE 1 Pedunculated FNH cases reported in the literature |
|----------------------------------------------------------|
| Authors | Year of publication | Gender | Age (years) | Symptoms | Size (cm) | Location | Treatment modality |
|---------|---------------------|--------|-------------|----------|----------|----------|-------------------|
| Brouquet et al13 | 1985 | Female | 40 | Asymptomatic | Unknown | Seg 4 | Surgery |
| Bader et al14 | 2001 | Female | - | Upper abdominal pain | 4.5 | Seg 5 | Surgery |
| Byrnes et al15 | 2004 | Female | 30 | Right upper quadrant pain | 3.5 | Seg 3 | Surgery |
| Wasif et al16 | 2008 | Female | 48 | Right upper quadrant pain | 3.2 | Seg 4 | Laparoscopic resection |
| Khan et al17 | 2011 | Female | 29 | Mass in the right upper quadrant | 9.5 | Seg 3 | Surgery |
| Badea et al4 | 2014 | Female | 29 | Pain in the right hypochondrium | 8 | Seg 5 | Laparoscopic resection |
| Zeina et al8 | 2016 | Female | 25 | Epigastric pain diarrhea | 4.8 | Seg 2 | Conservative management |
| Tsukui et al12 | 2017 | Male | 39 | Asymptomatic | 3.9 | Seg 6 | Laparoscopic resection |
| Martiniuc et al18 | 2018 | Female | 40 | Asymptomatic interhepatic-gastric mass | 5 | Seg 3 | Surgery |
| Our case | 2020 | Female | 38 | Asymptomatic | 4.5 | Seg 3 | Surgery |

4 | CONCLUSION

FNH is a common benign liver tumor. Pedunculated FNH, however, is a rare entity of a challenging diagnosis, since it may be mistaken for other benign or malignant liver tumors. This atypical presentation mandates complete surgical resection for definitive diagnosis and to avoid potential complications such as torsion of the pedicle or compression of the surrounding organs.

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CONFLICT OF INTEREST
Authors declare no conflict of interest.
AUTHOR’S CONTRIBUTION
Dr Ben Ismail Imen: involved in study concepts and manuscript writing. Dr Zenaidi Hakim: helped in data interpretation and manuscript evaluation. Dr Rebii Saber: involved in data acquisition. Dr Zoghlami Ayoub: critically revised the manuscript.

ETHICAL APPROVAL
Ethical approval was not required, and patient identifying knowledge was not presented in the report.

DATA AVAILABILITY STATEMENT
The data that support the findings of this study are available from the corresponding author upon reasonable request.

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