Sclerosing Hepatic Hemangioma Can Be Difficult to Differentiate from Liver Metastasis of Rectal Cancer: A Case Report

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Introduction: Sclerosing hepatic hemangiomas are a rare form of cavernous hemangioma, reported in 0.2% of autopsy cases. Preoperative diagnosis is difficult because of the variety of imaging findings. Herein, we report a case of hepatic sclerosing hemangioma that was difficult to differentiate from a liver metastasis of rectal cancer.

Case presentation: A 67-year-old man visited our hospital with a chief complaint of bleeding during defecation, and a colonoscopy revealed advanced rectal cancer. A dynamic contrast-enhanced magnetic resonance imaging (MRI) showed a 15 mm-sized tumor in S7 of the liver. In the arterial phase, the tumor interior showed low signal intensity, and the tumor margins were enhanced. The tumor interior was gradually stained from portal to equilibrium phases. Partial S7 resection was performed for liver metastasis from rectal cancer. Hematoxylin and Eosin staining revealed flattened endothelial cells with poor atypia that formed a lumen. Immunohistochemical staining was positive for CD31 and CD34, and the final diagnosis was sclerosing hemangioma.

Conclusion: Although a rare tumor, hepatic sclerosing hemangioma should always be considered as a differential diagnosis for liver tumors. If the diagnosis is difficult to make and malignancy cannot be ruled out, resection may be indicated as a diagnostic treatment.

Key words: Sclerosing hepatic hemangioma; liver metastasis; rectal cancer

INTRODUCTION

Cavernous hepatic hemangiomas are common benign liver tumors. By contrast, sclerosing hemangiomas represent a very rare form of hemangiomas that were reported to be present in only 0.2% of cases, in a study of 1,000 autopsies [1]. Hepatic hemangiomas become sclerosing hemangiomas through fibrosis, thrombosis, and other forms of degeneration, over long periods [2, 3]. Because they can present a variety of forms on imaging studies, they are often difficult to differentiate from malignant tumors, and may therefore be resected [4, 5]. Herein, we report a case of hepatic sclerosing hemangioma that was difficult to differentiate from a liver metastasis of rectal cancer.

CASE REPORT

A 67-year-old man visited a hospital with a chief complaint of bleeding during defecation, and a colonoscopy revealed advanced rectal cancer (Fig. 1). The patient was then referred to our hospital. He had no relevant medical or family history. He had consumed three cans of beer and smoked 20 cigarettes per day for 35 years. He had quit smoking 10 years prior to his presentation to hospital.

No abnormalities were observed on physical examination. Blood biochemistry showed no abnormal findings except for mild liver dysfunction (GOT: 34 U/L; GPT: 47 U/L) and mildly elevated glucose levels (126 mg/dL). His levels of other tumor markers were within normal limits (carcinoembryonic antigen: 3.8 U/mL; cancer antigen 19-9: 19.1 mg/mL). Additional imaging studies were performed to search for distant metastases. Abdominal ultrasonography revealed a 17 × 17 mm round hypoechoic mass in segment 7 of the liver (Fig. 2). Doppler examination revealed no blood flow inside the tumor. Abdominal contrast-enhanced computed tomography showed an indistinct bordering mass, 15 mm in diameter, in segment 7 of the liver, with a slightly lower density than the liver parenchyma (Fig. 3). Abdominal magnetic resonance imaging (MRI) revealed a low signal on fat-suppressed T1-weighted images, and a slightly higher signal on T2-weighted images (Fig. 4A, B). Abdominal dynamic contrast-enhanced MRI showed that the tumor had a low signal intensity before contrast enhancement, and showed a high signal intensity at the tumor periphery during the arterial phase. From the portal phase to the equilibrium phase, the inside of the tumor showed gradually staining. In the equilibrium phase, most ar-

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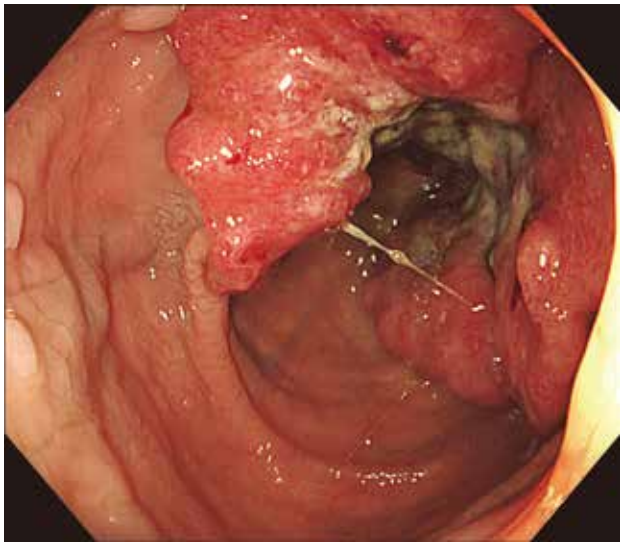

Fig. 1 Lower gastrointestinal endoscopy revealed a type 2 lesion in the rectum. Histopathology revealed a diagnosis of adenocarcinoma.

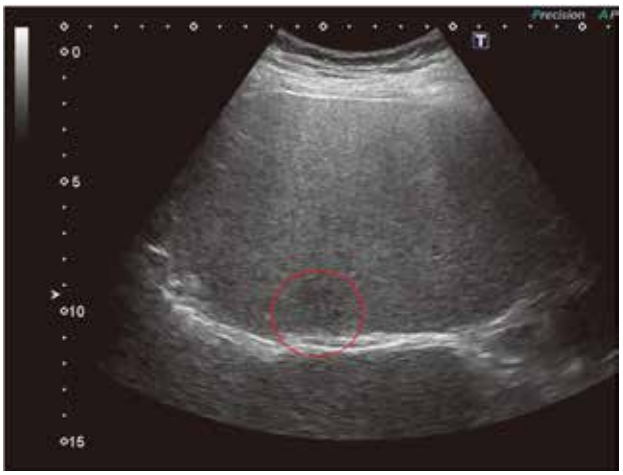

Fig. 2 Abdominal ultrasonography showing a 17 x 17 mm, round hypoechoic mass with indistinct borders and a heterogeneous interior in segment 7 of the liver (red circle).

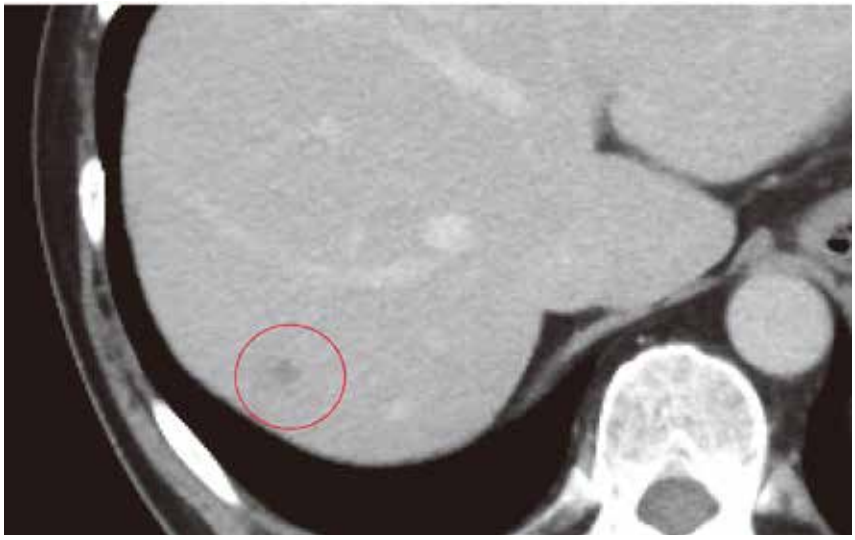

Fig. 3 Abdominal contrast-enhanced computed tomography showing a 15 mm-sized hypodense mass with indistinct borders and slightly contrasted margins in segment 7 of the liver (red circle).

eas of the tumor showed isointense signal with the liver parenchyma (Fig. 4C–F). Diffusion-weighted images showed high signal intensity, and a Gd-EOB-DPTA-enhanced MRI (EOB-MRI) of the hepatocellular phase showed a slightly lower signal than the liver parenchyma (Fig. 4G, H). The possibility that this liver mass was a metastasis from rectal cancer could not be ruled out — and, in the end, the patient was diagnosed with stage 4 rectal cancer. A laparoscopic anterior resection was performed to treat the primary rectal cancer. After three courses of Capecitabine therapy, we decided to perform radical resection of the single liver metastasis, because its size did not change significantly and there were no new lesions.

At the time of writing this report, our department is actively performing laparoscopic hepatectomy procedures; however, at the time of the patient's surgery this procedure was just being introduced. As S7 resection is a difficult operation, partial S7 resection was performed via laparotomy. The operation time was 258 min, and the total blood loss was 1,300 mL. Blood transfusions were not needed. The surgical specimen showed a grayish-white nodule measuring 15 × 17 mm (Fig. 5). Hematoxylin and Eosin staining revealed flattened endothelial cells with poor atypia that formed a lumen and proliferated in the lesion. The mass was rich in acidophilic interstitial components and partially edematous. No capsular formation was observed around it, and inflammatory cell infiltration was noticeable along its border with the surrounding normal liver tissue (Fig. 6A, B). Immunohistochemical staining of the tumor showed that the proliferating endothelial cells were positive for CD31 and CD34 and negative for D2–40, with no evidence of adenocarcinoma. Based on these findings, the final diagnosis was sclerosing hepatic hemangioma

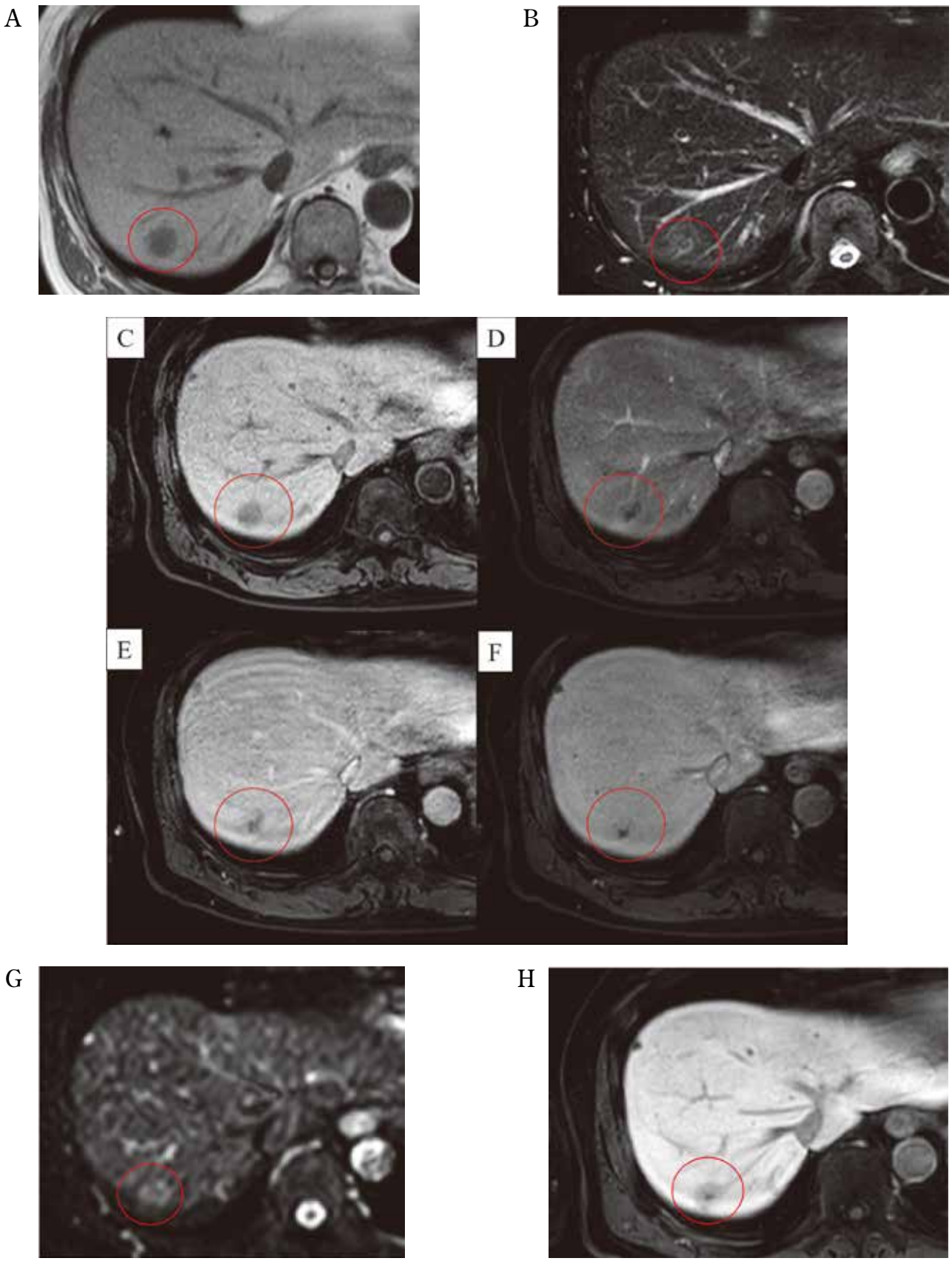

Fig. 4 (A) T1-weighted magnetic resonance imaging: the tumor showed a lower signal compared to the liver parenchyma (red circle). (B) T2-weighted magnetic resonance imaging: the tumor showed a slightly higher signal than the liver parenchyma (red circle). (C–F) These images are the findings of a dynamic contrast-enhanced MRI. (C) In the pre-contrast images showed low signal entire the tumor. (D) In the arterial phase, the tumor margins showed high signal intensity. (E, F) The tumor interior gradually stained from the portal phase to the equilibrium phase. In the equilibrium phase, most area of the tumor was isointense with the liver parenchyma. (G) Diffusion-weighted magnetic resonance imaging: the tumor showed a high signal (red circle). (H) Gd-EOB-DPTA-enhanced magnetic resonance hepatocellular phase: the tumor showed a slightly low signal than the liver parenchyma (red circle).

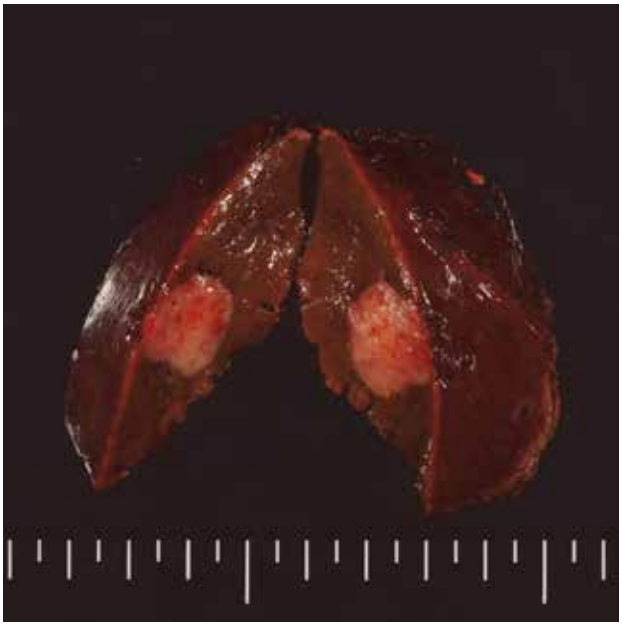

Fig 5 The surgical specimen: a 15 x 17 mm hard, grayish-white nodule was observed in the resected liver.

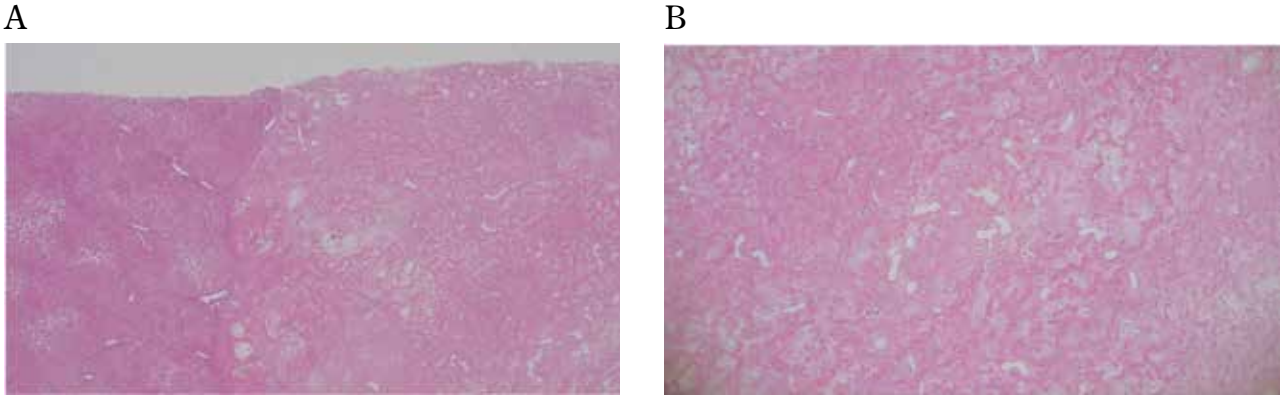

Fig. 6 Pathological findings: (A) The border between the tumor and normal surrounding liver tissue was clear. Within the tumor, there was an edematous thick stroma and deposits of eosinophilic material. (Hematoxylin and Eosin; magnification, × 20) (B) Numerous small vessels are present in the tumor. (Hematoxylin and Eosin; magnification, × 40)

(Fig. 7A, B). There were no postoperative complications, and the patient was discharged on the 13th postoperative day. As of 42 months postoperatively, there has been no recurrence of the rectal cancer or any new liver tumors.

DISCUSSION

Hepatic hemangiomas are benign tumors that are often found incidentally in the liver. They are reported to be more common in women in their 30s and 50s [6]. They can be classified as capillary, cavernous, or sclerosing hemangiomas [7]. Patients are typically monitored through regular follow-ups, but large cavernous hemangiomas may rupture or cause disseminated intravascular coagulation, at which point they should be treated surgically [8, 9]. Sclerosing hemangiomas were first reported by Shepherd *et al.* in 1983 [10], and their frequency is extremely low [1]. The clinical course is variable and may be diagnosed using imaging studies and clinical symptoms such as abdominal pain [11].

The typical findings on dynamic contrast-enhanced CT scans of hepatic hemangiomas include early staining of the tumor margins in the arterial phase and spread of the contrast effect to the tumor center in the portal or delayed phases [12, 13]. Some hemangiomas are iso-absorbed by the liver in the portal and delayed phases; however, once stained, the staining does not become hypo-absorbed, which is a useful finding for differentiating hemangiomas from hepatocellular carcinomas and other tumors [14, 15] However, sclerosing hemangiomas are very difficult to diagnose, because they often do not show the typical imaging findings of hemangiomas. This is due to degeneration, such as fibrosis and thrombus formation inside the tumor, resulting in a variety of contrast patterns. Unfortunately, dynamic contrast-enhanced CT was not performed in this case, and the contrast pattern was not evaluated.

Typical MRI findings of hemangiomas include uniformly high signal intensity on T2-weighted images and uniformly low signal intensity on T1-weighted

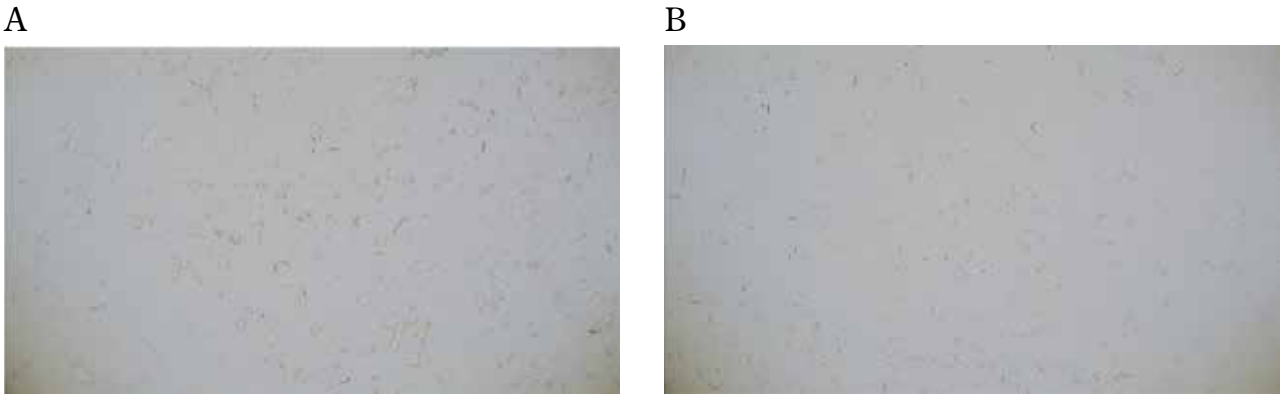

Fig. 7 Immunohistochemistry: proliferating endothelial cells are positive for CD31 (A) and CD34 (B).

images [16]. In our case, T1-weighted images showed a uniform low signal, but T2-weighted ones showed a heterogeneous pale high signal. EOB-MRI showed that the tumor was slightly lower in signal than the liver parenchyma. These findings are not typical MRI features of hemangiomas. In addition, this case also showed a high signal on diffusion MRI. Miyata *et al.* reported that the apparent diffusion coefficient (ADC) mean value on diffusion MRI was significantly higher in hepatic sclerosing hemangiomas than in existing malignant tumors, which represent a useful feature in terms of preoperative differentiation [3].

Treatment for sclerosing hemangiomas is typically administered during follow-up if a preoperative diagnosis of hemangioma can be made. However, in a number of reports (as well as the present case) the tumor was resected due to the difficulty of differentiating it from a malignancy, and a definitive diagnosis was reached only through histopathological examination [4, 5, 17–20]. Wakasugi *et al.* reported a case of multiple resected hepatic sclerosing hemangiomas that were difficult to diagnose. These were resected because of a lack of typical imaging findings and a history of gastric carcinoids in the patient, suggesting that multiple cases are more difficult to differentiate from metastatic liver tumors [5].

Based on the morphological findings of the mass excised in this case, the specimen was a hard white nodule that was similar to an adenocarcinoma, making it difficult to identify as a hemangioma. Preoperative biopsy may be effective in some cases, but it is difficult to perform given the possibility of contributing to peritoneal dissemination in the case of malignant tumors (e.g., cholangiocarcinomas), and the possibility of not obtaining sufficient tissue in the case of hard, vitreous-degenerated tumors. In one study, ablation was performed; however, the tumor was hard, rendering this approach ineffective, so resection was ultimately performed [4]. In the present case, liver resection was performed via laparotomy. Recently, laparoscopic liver resection techniques have advanced and become more widespread, with reported advantages such as decreased intraoperative blood loss and shorter hospital stays [21, 22]. It has also been reported that there is no difference in the long-term prognosis between malignancy and open resection [21, 22]. Minimally invasive laparoscopic liver resection for diagnostic therapy is expected to become the treatment of choice for liver tumors that are difficult to distinguish from malignant ones, as in the present case.

CONCLUSION

We encountered a case of hepatic sclerosing hemangioma comorbid with rectal cancer that was difficult to distinguish from a metastatic tumor. Although rare, hepatic sclerosing hemangioma should always be considered as a differential diagnosis for liver tumors. If the diagnosis is difficult to make and the tumor is resectable, resection can be considered as a diagnostic therapy.

INFORMED CONSENT

Informed consent was obtained from the patient for the submission of this report, in accordance with the COPE guidelines.

REFERENCES

1) Berry CL. Solitary “necrotic nodule” of the liver: a probable pathogenesis. J Clin Pathol 1985; 38: 1278–80.
2) Koyama R, Minagawa N, Maeda Y, Shinohara T, Hamada T. A hepatic sclerosing hemangioma emerged in the postoperative course of multiple gastric carcinoid tumors masquerading as metachronous liver metastasis. Int J Surg Case Rep 2019; 58: 1–5.
3) Miyata T, Beppu T, Kuramoto K, Nakagawa S, Imai K, Hashimoto D, *et al.* Hepatic sclerosed hemangioma with special attention to diffused-weighted magnetic resonance imaging. Surg Case Rep 2018; 4: 3.
4) Xu L, Yang X, Ke S, Ding XM, Wang SH, Gao J, *et al.* Resection as first-line therapy for large hepatic sclerosing hemangioma: a case report. Onco Targets Ther 2019; 12: 6839–3842.
5) Wakasugi M, Ueshima S, Tei M, Tori M, Yoshida Y, Tsujimoto M, *et al.* Multiple hepatic sclerosing hemangioma mimicking metastatic liver tumor successfully by laparoscopic surgery: Report of a case. Int J Surg Case Rep 2015; 8: 137–140.
6) Cseiro-Alves F, Brito J, Araujo AE, Belo-Soares P, Rodrigues H, Cipriano A, *et al.* Liver hemangioma: common and uncommon findings and how to improve the differential diagnosis. Eur Radiol 2007; 20: S49–60.
7) Bajenaru N, Balaban V, Săvulescu F, Campeanu I, Patrascu T. Hepatic hemangioma-review-. J med Life 2015; 8: 4–11.
8) Hanazaki K, Kajiwara S, Matsushita A, Monma T, Koide N, Nimura Y, *et al.* Hepatic resection of giant giant cavernous hemangioma of the liver. J Clin Gastroenterol 1999; 29: 257–60.
9) Donati M, Stavrou GA, Donati A, Oldhafer KJ. The risk of spontaneous rupture of liver hemangiomas: a critical review of the literature. J Hepatobilialy Sci 2011; 6: 797–805.

10) Shepherd NA, Lee G. Solitary necrotic nodules of the liver simulating hepatic metastasis. J Clin Pathol 1983; 36: 1181–1183.
11) Aziz H, Brown ZJ, Baghdadi A, Kamel IR, Pawlik TM. A Comprehensive Review of Hepatic Hemangioma Management. J Gastrointest Surg 2022; 26: 1998–2007.
12) Ashida C, Fishman EK, Zerhouni EA, Herlong FH, Siegelman SS. Computed tomography of hepatic cavernous hemangioma. J Comput Aissist tomogr 1987;11: 455–460.
13) Freeny PC, Marks WM. Hepatic hemangioma: dynamic bolus CT. AJR Am J Roentgenol 1986; 147: 711–9.
14) Hanafusa K, Ohnishi I, Himeno Y, Suzuki S, Shibuya H. Hepatic hemangioma. : findings with two-phase CT. Radiology 1995; 196: 465–9.
15) Hanafusa K, Ohashi I, Gomi N, HimenoY, Wakita T, Shibuya H. Differential diagnosis of early homogeneously enhancing hepatocellular carcinoma and hemangioma by two-phase CT. J Comput Aissist tomogr 1997; 21: 361–8.
16) Semelka RC, Brown ED, Ascher SM, Patt RH, Bagley AS, Li W, *et al*. Hepatic hemangioma: a multi-institutional study of appearance on T2-weighted and serial gadolinium-enhanced gradient-echo MR images. Radiology 1994; 192: 401–6.
17) Yamada S, Shimada M, Utsunomiya T, Morine Y, Imura S, Ikemoto T, *et al*. Hepatic screlosed hemangioma which was misdiagnosed as metastasis of gastric cancer: report of a case. J Med Invest 2012; 59; 270–274.
18) Yamashita Y, Shimada M, Taguchi K, Gion T, Hasegawa H, Utsunomiya T, *et al*. Hepatic sclerosing hemangioma mimicking a metastatic liver tumor: report of a case. Surg Today 2000; 30: 849–852.
19) Aibe H, Honda H, Kuroiwa T, Yoshimitsu K, Irie H, Tajima T, *et al*. Sclerosed hemangioma of the liver. Abdom Imaging 2001; 26: 496–499.
20) Lauder C, Garcea G, Kanhere H, Maddern GJ. Sclerosing hemangiomas of the liver.: two cases of mistaken identity. HPB surg 2009; 2009: 473591.
21) Yoon YI, Kim KH, Cho HD, Kwon JH, Jung DH, Park GC, *et al*. Long-term perioperative outcomes of pure laparoscopic liver resection versus open liver resection for hepatocellular carcinoma: a retrospective study. Surg Eondosc 2020; 34: 796–805.
22) Nicolás M, Czerwonko M, Ardiles V, Sánchez Claria R, Mazza O, de Santibañes E, *et al*. Laparoscopic vs open liver resection for metastatic colorectal cancer: analysis of surgical margin status and survival. Langenbecks Arch Surg 2022; 407: 1113–1119.