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OPINION REVIEW

- 608** Is precision medicine for colorectal liver metastases still a utopia? New perspectives by modern biomarkers, radiomics, and artificial intelligence

Viganò L, Jayakody Arachchige VS, Fiz F

MINIREVIEWS

- 624** Recent advances in the diagnostic evaluation of pancreatic cystic lesions

Ardeshta DR, Cao T, Rodgers B, Onongaya C, Jones D, Chen W, Koay EJ, Krishna SG

ORIGINAL ARTICLE**Basic Study**

- 635** Effects of viremia and CD4 recovery on gut “microbiome-immunity” axis in treatment-naïve HIV-1-infected patients undergoing antiretroviral therapy

Russo E, Nannini G, Sterrantino G, Kiros ST, Pilato VD, Coppi M, Baldi S, Niccolai E, Ricci F, Ramazzotti M, Pallecchi M, Lagi F, Rossolini GM, Bartoloni A, Bartolucci G, Amedei A

Case Control Study

- 653** Atrophic gastritis and gastric cancer tissue miRNome analysis reveal hsa-miR-129-1 and hsa-miR-196a as potential early diagnostic biomarkers

Varkalaite G, Vaitkeviciute E, Inciuraite R, Salteniene V, Juzenas S, Petkevicius V, Gudaityte R, Mickevicius A, Link A, Kupcinskas L, Leja M, Kupcinskas J, Skieceviciene J

Retrospective Study

- 665** Validation of the PAGE-B score to predict hepatocellular carcinoma risk in caucasian chronic hepatitis B patients on treatment

Gokcen P, Guzelbulut F, Adali G, Degirmenci Salturk AG, Ozturk O, Bahadir O, Kanatsiz E, Kiyak M, Ozdil K, Doganay HL

CASE REPORT

- 675** Gallbladder Burkitt's lymphoma mimicking gallbladder cancer: A case report

Hosoda K, Shimizu A, Kubota K, Notake T, Hayashi H, Yasukawa K, Umemura K, Kamachi A, Goto T, Tomida H, Yamazaki S, Narusawa Y, Asano N, Uehara T, Soejima Y

LETTER TO THE EDITOR

- 683** COVID-19, liver dysfunction and pathophysiology: A conceptual discussion

Gonçalves Júnior J

- 689** Comments on validation of conventional non-invasive fibrosis scoring systems in patients with metabolic associated fatty liver disease

Hong JG, Yan LJ, Li X, Yao SY, Su P, Li HC, Ding ZN, Wang DX, Dong ZR, Li T

ABOUT COVER

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CASE REPORT

Gallbladder Burkitt's lymphoma mimicking gallbladder cancer: A case report

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Abstract

BACKGROUND

Malignant lymphoma is a rare form of gallbladder malignancy. Most of these malignancies are diffuse large B-cell lymphomas or mucosa-associated lymphoid tissue-type lymphomas; however, Burkitt's lymphoma of the gallbladder is extremely rare, and only two previous reports are available in the literature. Herein, we report a rare case of Burkitt's lymphoma of the gallbladder mimicking gallbladder adenocarcinoma.

CASE SUMMARY

An 83-year-old man with no abdominal complaints was found to have a gallbladder tumor and periportal lymph node enlargement on computed tomography (CT) performed for hypertension screening. His laboratory data revealed slightly elevated serum levels of carcinoembryonic antigen and soluble interleukin 2 receptor. Imaging examinations revealed two irregular and contrast-enhanced masses extending into the gallbladder lumen, but these did not infiltrate the serosa. Moreover, a periportal lymph node had enlarged to 30 mm. Based on these findings, we diagnosed the patient as having gallbladder adenocarcinoma with lymph node metastasis, which was treated using bile duct resection with gallbladder bed resection and periportal lymph node dissection. However, the patient was finally diagnosed as having Burkitt's lymphoma. Although the surgical margin was pathologically negative, recurrence was noted at the hepatic

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radical margin and superior pancreaticoduodenal lymph nodes on positron emission tomography/CT soon after discharge. Thus, he was referred to a hematologist and started receiving treatment with reduced-dose cyclophosphamide, doxorubicin, vincristine, and prednisone.

CONCLUSION

Burkitt's lymphoma can occur in the gallbladder. Biopsy can be useful in cases with findings suggestive of gallbladder malignant lymphoma.

Key Words: Gallbladder; Malignant lymphoma; Burkitt's lymphoma; Gallbladder cancer; Lymphadenopathy; Case report

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Core Tip: Malignant lymphoma is a rare form of gallbladder malignancy, and Burkitt's lymphoma of the gallbladder is especially rare, with only two previous reports available in the literature. We report a case of gallbladder Burkitt's lymphoma that was preoperatively indistinguishable from gallbladder carcinoma. Unfortunately, the patient had lymphoma recurrence immediately after the surgery because of delayed chemotherapy initiation owing to postoperative complications due to an extended surgery. Although accurate preoperative diagnosis of gallbladder malignant lymphoma is quite difficult, some findings are suggestive of gallbladder malignant lymphoma, and hence, biopsy is recommended in these cases.

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INTRODUCTION

Malignant lymphoma of the gallbladder is extremely rare[1-4]. In almost all cases of this malignancy, patients are diagnosed as having gallbladder adenocarcinoma or cholecystitis at the time of surgery, and preoperative diagnosis is extremely difficult. Although several reports have documented malignant lymphomas of the gallbladder, most of these malignancies are diffuse large B-cell lymphomas and mucosa-associated lymphoid tissue-type lymphomas (MALTomas)[2,4,5], and only two reports have previously documented Burkitt's lymphoma of the gallbladder[6,7]. Herein, we report the case of a patient with Burkitt's lymphoma of the gallbladder mimicking gallbladder adenocarcinoma.

CASE PRESENTATION

Chief complaints

An 83-year-old man with no abdominal complaints was found to have a gallbladder tumor along with periportal lymph node enlargement and was admitted to our institution for further investigation.

History of present illness

The tumor was detected during contrast-enhanced computed tomography (CT) performed for a detailed examination of hypertension.

History of past illness

The patient had a history of cerebral artery stenosis and paroxysmal atrial fibrillation.

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Personal and family history

No personal and family history.

Physical examination

Abdominal examination revealed no palpable mass or tenderness.

Laboratory examinations

Laboratory examinations performed upon admission revealed mild to moderate renal dysfunction (creatinine: 1.21 mg/dL) and slightly elevated levels of serum carcinoembryonic antigen and serum soluble interleukin 2 receptor (6.1 ng/mL and 635 IU/mL, respectively). Other laboratory data were within normal limits.

Imaging examinations

A contrast-enhanced abdominal CT scan revealed two irregular and highly contrast-enhanced masses at the neck and body of the gallbladder as well as periportal lymph node enlargement, measuring 30 mm × 20 mm in diameter and consistent with gallbladder cancer lymph node metastasis (Figure 1). Magnetic resonance imaging revealed that the tumor signal was hypointense on T1-weighted imaging and hyperintense on T2-weighted and diffusion-weighted imaging (Figure 2). Positron emission tomography (PET) revealed increased ¹⁸F-fluorodeoxyglucose uptake in the tumor (Figure 3). Endoscopic ultrasonography (EUS) showed a heterogeneous echoic mass extending into the lumen, but it did not infiltrate the serosa (Figure 4).

FINAL DIAGNOSIS

The final diagnosis was gallbladder adenocarcinoma with lymph node metastasis.

TREATMENT

On the basis of these findings, the patient underwent bile duct resection with gallbladder bed resection and periportal lymph node dissection. The surgical findings revealed the gallbladder tumor was relatively softer than ordinary gallbladder cancers. The swollen lymph node was firm but did not invade the portal vein. However, the patient developed severe aspiration pneumonia and bile leakage after the surgery, which were treated conservatively. The patient was discharged 2 mo after the surgery.

OUTCOME AND FOLLOW-UP

Histologic examination revealed periportal lymphadenopathy and two tumors at the neck and body of the gallbladder, measuring 27 mm × 20 mm and 20 mm × 18 mm in diameter, respectively. Histological findings also showed monotonous lymphoid cells with hemophagocytosis by macrophages. Immunohistochemical staining for markers showed the presence of CD10, BCL6, and c-Myc and the absence of BCL2. The Ki-67 index was > 80% (Figure 5). Therefore, the patient was finally diagnosed as having Burkitt's lymphoma. Although the surgical margin was pathologically negative, recurrence was noted at the hepatic radical margin and superior pancreaticoduodenal lymph nodes on PET-CT immediately after discharge. Thus, he was referred to a hematologist and started receiving treatment with reduced-dose cyclophosphamide, doxorubicin, vincristine, and prednisone.

DISCUSSION

Malignant lymphoma of the gallbladder is a rare form of gallbladder malignancy, which accounts for 0.1%-0.2% of all gallbladder cancers[1-3]. In previous reports, most of these malignancies were documented to be diffuse large B-cell lymphomas and MALTomas[2,4], and only two reports had documented Burkitt's lymphoma of the gallbladder[6,7]. Compared to previous cases, the present case yielded some interesting clinical and imaging findings.

Table 1 Diagnostic criteria for gastrointestinal malignant lymphoma

| Ref. | Diagnostic criteria |
|-----------------|---|
| Dawson et al[8] | Absence of palpable superficial lymphadenopathy Absence of obvious enlargement of mediastinal lymph nodes Normal level of total and differential white blood cell counts The bowel lesion predominating and the only lymph node obviously affected being those in its immediate neighborhood Absence of tumor in the liver and spleen |
| Lewin et al[9] | Exhibiting gastrointestinal symptoms or predominant lesions in the gastrointestinal tract |

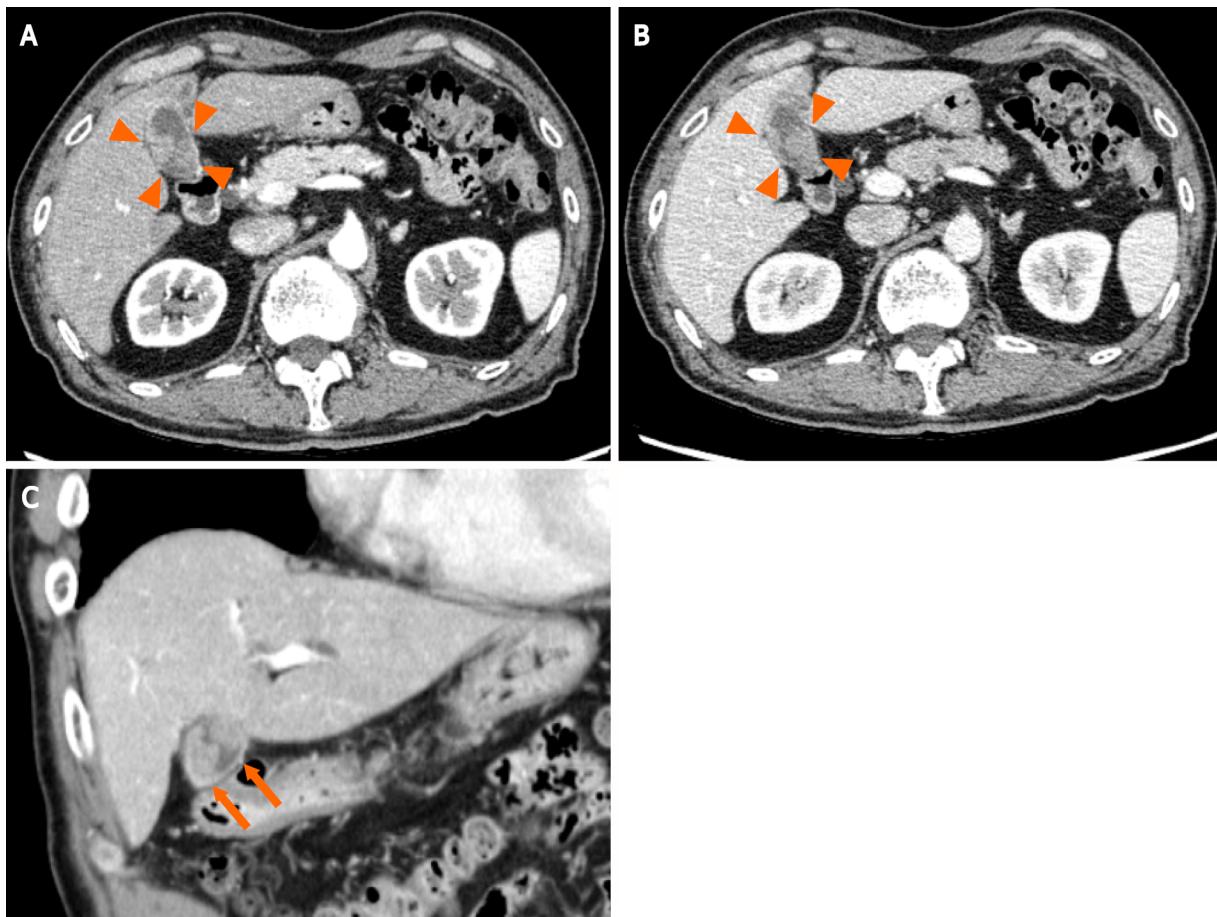


Figure 1 A contrast-enhanced abdominal computed tomography scan shows two irregular and highly contrast-enhanced masses (arrowheads and arrow) at the neck and body of the gallbladder as well as periportal lymph node enlargement, which is consistent with gallbladder cancer lymph node metastasis. A: Axial section image in the early phase showing neck of the gallbladder; B: Axial section image in the delayed phase showing neck of the gallbladder in the delayed phase; C: Coronal sectional image showing body of the gallbladder.

First, this is potentially the first reported case of “primary” gallbladder Burkitt’s lymphoma. According to the diagnostic criteria of primary gastrointestinal lymphoma (**Table 1**) defined by Dawson et al[8] and Lewin et al[9], the previous two cases were diagnosed as “secondary” gallbladder Burkitt’s lymphoma because they included extra-gallbladder lesions, such as duodenal, hepatic, or central nervous system lesions. In contrast, in the present case, the tumor was localized in the gallbladder and a periportal lymph node. Although no gastrointestinal symptoms were observed because the primary site was the gallbladder, other criteria were fulfilled. Therefore, the patient was diagnosed as having “primary” gallbladder Burkitt’s lymphoma, and the case was considered novel.

Second, gallbladder Burkitt’s lymphoma can present as a localized disease that mimics gallbladder cancer. The imaging features of malignant lymphoma were reported by Ono et al[3], who reported that high-grade malignant lymphomas showed

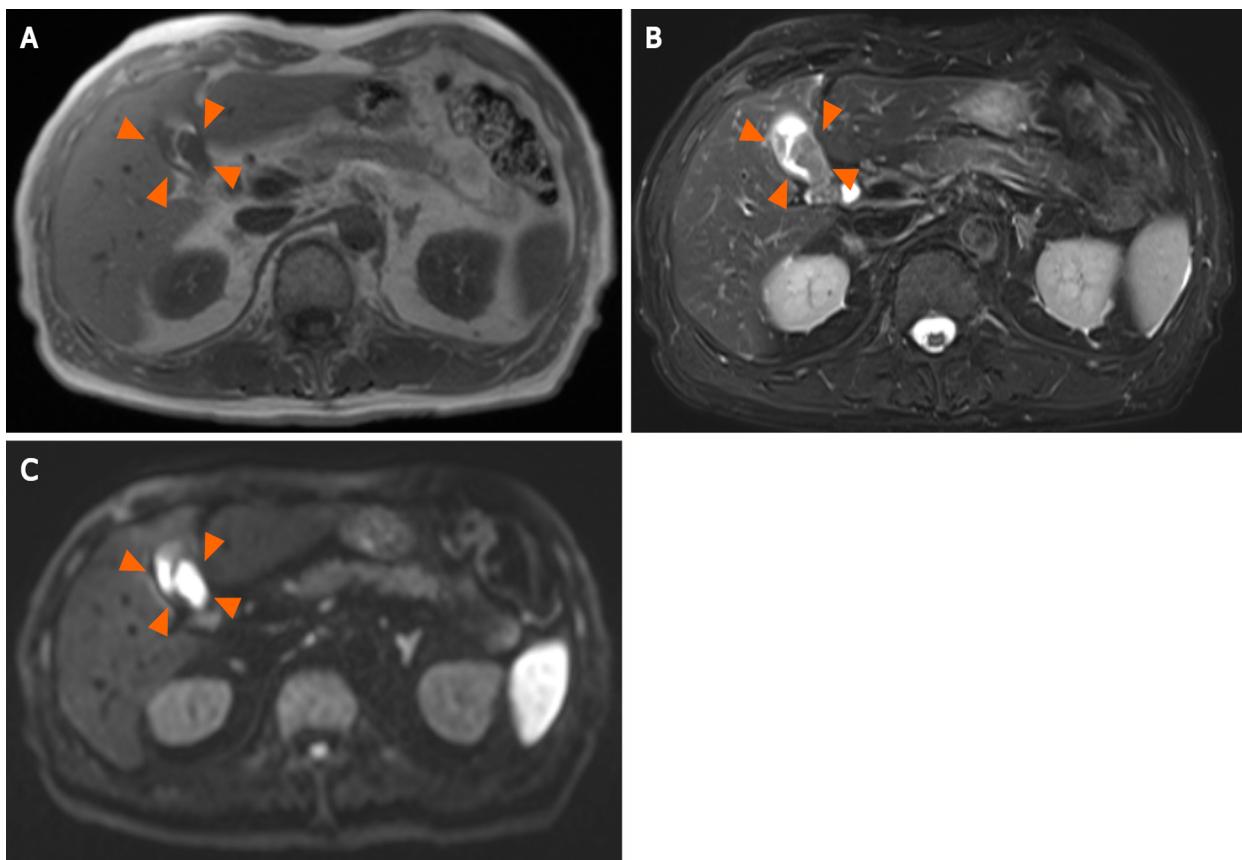


Figure 2 Magnetic resonance imaging reveals a hypointense tumor signal. A: T1-weighted imaging (arrowheads); B: A hyperintense signal on T2-weighted imaging (arrowheads); C: Diffusion-weighted imaging (arrowheads).



Figure 3 Endoscopic ultrasonography shows a heterogeneous echoic mass (arrows) with internal partially low echo (arrowheads). The mass extends into the lumen but does not infiltrate the serosa.

solid and bulky masses or irregular wall thickening. In addition, because Burkitt's lymphoma is a highly aggressive and rapidly progressive disease, some extranodal sites are generally involved at the time of diagnosis[10] and tumor localization around the primary lesion is rare[11]. In this case, the malignancy was localized in the body of the gallbladder and a periportal lymph node. Since these findings were consistent with the imaging findings of gallbladder adenocarcinoma, we could not confirm a preoperative diagnosis of malignant lymphoma.

Unfortunately, the patient had lymphoma recurrence 2 mo after the surgery because the introduction of chemotherapy had to be delayed owing to postoperative complications. Burkitt's lymphoma is a highly aggressive disease, but it is highly sensitive to chemotherapy. Therefore, chemotherapy has the highest priority in the treatment of

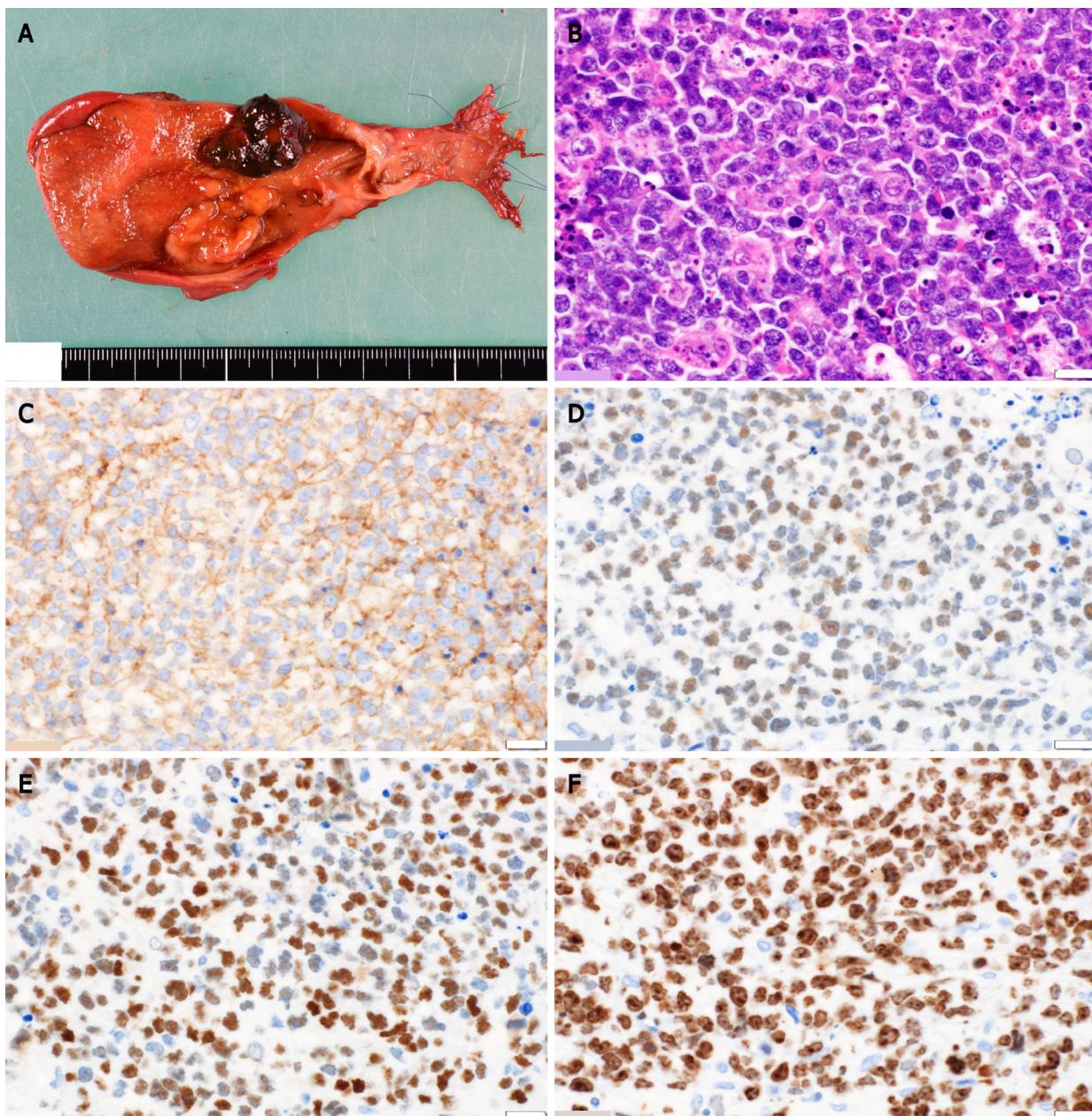


Figure 4 Histologic examination. A: Periportal lymphadenopathy and two tumors at the neck and body of the gallbladder, measuring 27 mm × 20 mm and 20 mm × 18 mm in diameter, respectively; B: Histological findings reveal monotonous lymphoid cells with hemophagocytosis by macrophages; C-E: Immunohistochemical staining for markers shows the presence of CD10 (C), BCL6 (D), and c-Myc (E) and the absence of BCL2; F: The Ki-67 index is > 80%. The white scale bars represent 1 mm.

Burkitt's lymphoma[10], and we should have introduced chemotherapy as soon as possible after the surgery. If we had been aware of the possibility of gallbladder malignant lymphoma, we could have avoided the extended procedure and could have initiated chemotherapy at the appropriate time.

Although the accurate preoperative diagnosis of gallbladder malignant lymphoma is quite difficult[2,12], some previous reports[3,12,13] have suggested the possibility of a precise preoperative diagnosis based on imaging findings. Specifically, Ono *et al*[3] showed that the signal intensity of the gallbladder wall on T2-weighted imaging is more hypointense in malignant lymphoma than in carcinoma. In addition, Kato *et al* [12] reported the usefulness of an internal partially low echo in distinguishing malignant lymphoma from carcinoma. On a retrospective review of the present case, although no obvious differences were observed in the intensity of T2-weighted imaging when compared to that of gallbladder cancers, EUS revealed an internal partially low echo of the tumor. Furthermore, in this case, a discrepancy existed in that no serosal invasion was observed despite the size of the tumor and presence of lymphadenopathy. We think this might be a characteristic finding that distinguishes

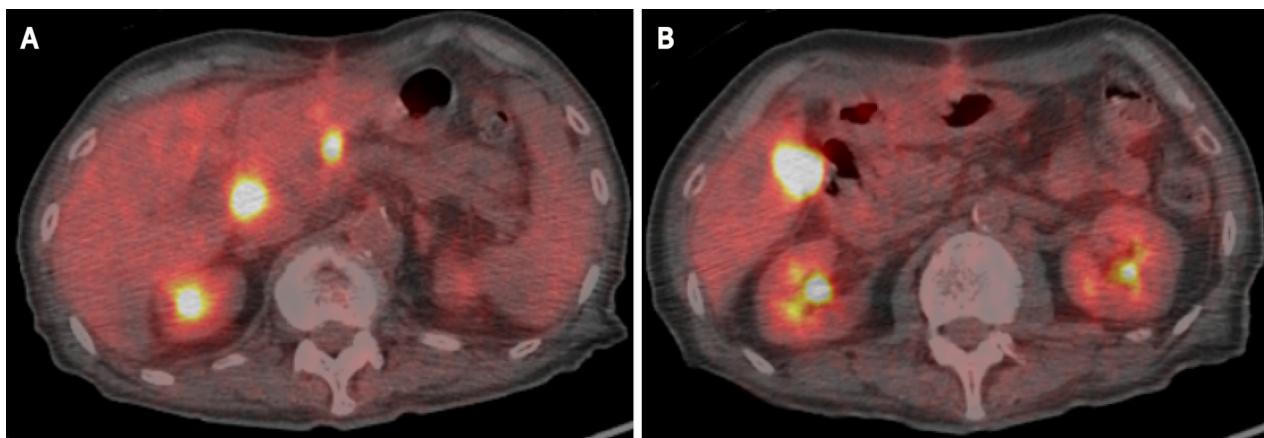


Figure 5 Positron emission tomography reveals increased ¹⁸F-fluorodeoxyglucose uptake at the superior pancreaticoduodenal lymph nodes and hepatic radical margin. A: Superior pancreaticoduodenal lymph nodes; B: Hepatic radical margin.

gallbladder malignant lymphoma from carcinoma.

Nevertheless, a biopsy examination seems the best diagnostic technique in the present case, considering the possibility of malignant lymphoma, and hence, biopsy should be planned for patients with the above imaging findings.

CONCLUSION

Burkitt's lymphoma can occur in the gallbladder. Therefore, this disease should be considered in the differential diagnosis of a gallbladder tumor, and biopsy can be useful in facilitating the early introduction of chemotherapy in cases with suggestive imaging findings.

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