Adenocarcinoma Arising in Segmental Adenomyomatosis of the Gallbladder: A Case Report

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We report here a rare case of adenocarcinoma arising from segmental adenomyomatosis of the gallbladder itself. CT images showed segmental annular wall thickening of the gallbladder body with regional lymph node enlargement, which could not exclude adenomyomatosis associated with malignancy. Therefore, we performed MRI and PET/CT for further evaluation. T2 weighted MRI revealed multiple tiny intramural cystic lesions in the focal wall thickening of the gallbladder, suggesting adenomyomatosis. However, PET/CT showed increased activity in the lesion of the gallbladder and regional lymph node, suggesting malignancy, which was pathologically confirmed as adenocarcinoma arising from adenomyomatosis.

Index words : Gallbladder Neoplasms
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Adenomyomatosis is a relatively common benign abnormality of the gallbladder, which has generally been found to have no risk of malignant transformation (1). However, a few recent cases have described carcinoma arising from adenomyomatosis of the gallbladder (1–5). To the best of our knowledge, a case of adenocarcinoma occurring in the segmental adenomyomatosis itself has not been reported in the radiologic literature. We report the computed tomographic (CT), magnetic resonance (MR), and positron emission tomographic (PET) findings correlated with the histopathologic features in a case of adenocarcinoma arising in segmental adenomyomatosis of the gallbladder itself.

Case Report

A 67-year-old man was admitted to our hospital with recurrent chest discomfort. The patient had a previous history of endoscopic submucosal dissection (ESD) due to early gastric cancer six months prior. A physical examination was found to be unremarkable.

The laboratory data were as follows: leukocyte count, $7.41 \times 10^3$/mm$^3$; ESR, 39 mm/h; total serum bilirubin, 1.34 mg/dL; AST, 337 IU/L; ALT, 202 IU/L; CA 19–9, 899.42 U/mL. The patient underwent an abdominal multi-detector computed tomography (MDCT) for a follow-up evaluation of ESD. The CT images showed segmental annular wall thickening in the body of the gallbladder with luminal narrowing, small amounts of sludge in the gallbladder, and enlargement of the common hepatic and left paraaortic lymph nodes (Figs. 1A, B). These findings suggested malignancy of the gallblad-
der with lymph node metastases, but we could not exclude adenomyomatosis. MR imaging for the differentiation of segmental adenomyomatosis and gallbladder cancer was performed and showed segmental annular wall thickening similar to those of the CT findings. Moreover, hyperintense Rokitansky-Aschoff sinuses in the form of multiple tiny intramural cystic lesions were readily identified on T2-weighted images, which were suggestive of gallbladder malignancy associated with segmental adenomyomatosis (Fig. 1C). Therefore, we performed PET with fluorine 18 fluorodeoxyglucose (FDG) for further evaluation. The PET/CT images showed increased activity in the gallbladder and an enlarged common hepatic lymph node, which was consistent with gallbladder malignancy and lymph node metastasis (Fig. 1D). Radical cholecystectomy and lymphadenectomy was performed for gallbladder malignancy. Pathologically, the gallbladder was 6.5 × 4 cm, and was contained in an encircling mass lesion [3.5 × 3 × 1.3 cm] with multiple tiny to small intramural cystic lesions (Fig. 1E). Microscopically, there was a proliferation of smooth muscle bundles around the sinuses as well as a thickened muscular layer, which were characteristic of segmental adenomyomatosis (Fig. 1F). Microscopically, some part of the mucosal epithelium was composed of papillary projections with cellular and

![Image](https://example.com/image1.png)

**Fig. 1.** Adenocarcinoma arising from segmental adenomyomatosis of the gallbladder in a 67-year-old man.
A. Transverse contrast-enhanced MDCT image showing focal annular wall thickening (arrow) of the gallbladder.
B. Transverse contrast-enhanced MDCT image showing an enlarged common hepatic lymph node (arrowhead), suggesting metastasis.
C. Transverse T2-weighted MR image showing multiple tiny intramural hyperintense cystic lesions (thin arrows), which are not identified on the CT image.
D. Transverse fused PET/CT image showing increased FDG activity in the annular wall thickening of the gallbladder (arrow) and common hepatic lymph nodes (arrowhead), which are consistent with gallbladder cancer and lymph node metastasis.
E. Photograph of the cut surface of the gross specimen shows a yellowish-white colored wall thickening with multiple tiny intramural cysts.
structural atypia, which was diagnosed as papillary adenoma. Some parts of the mucosal epithelium were diagnosed as adenocarcinoma, which exhibited enlarged and hyperchromatic nuclei with frequent mitoses. A transitional area was present between atypical cells and adenocarcinoma cells (Fig. 1F). The tumor invaded the perimuscular connective tissue of the gallbladder, but no invasion of the adjacent liver was noted at histopathologic examination. The immunohistochemical stain with p53 showed positive staining for adenocarcinoma (Fig. 1G).

Discussion

Adenomyomatosis is a fairly common benign lesion of the gallbladder that is present in more than 20% of patients who undergo a cholecystectomy for biliary symptoms [1].

Adenomyomatosis of the gallbladder is characterized by hyperplasia and hypertrophy of the tissues which constitute its wall. Epithelial cells undergo marked hyperplasia without other structural changes. The resulting epithelial proliferation forms invaginations and diverticula which penetrate the muscular layer and are called Rokitansky-Aschoff sinuses [6]. Adenomyomatosis is a hyperplasia of the gallbladder wall characterized by excessive proliferation of the Rokitansky-Aschoff sinuses and thickening of the fibromuscular layer (2). According to the extent of involvement, adenomyomatosis is classified into three types; [a] localized or fundal; [b] segmental or annular; and [c] diffuse. The localized type, also called adenomyoma, is usually found at the fundus of the gallbladder. The segmental type is typically located in the body of the gallbladder, giving it an hourglass configuration upon gross inspection. The diffuse type is a thickened wall throughout the entire gallbladder [7]. Pathologically, a Rokitansky-Aschoff sinus within the thickened muscular layer of the gallbladder is the characteristic finding of adenomyomatosis [6, 8]. Traditionally, adenomyomatosis has not been considered to pose the risk of malignancy [1]. Therefore, the depiction of the Rokitansky-Aschoff sinus should be important for the accuracy of radiologic diagnoses of adenomyomatosis [8].

Localized or segmental adenomyomatosis of the gallbladder may mimic gallbladder carcinomas (5). Therefore, radiologists should recognize the importance of radiologic differentiation between adenomyomatosis and gallbladder cancer. Abnormal gallbladder wall thickening and enhancement are common but nonspecific CT features of adenomyomatosis. However, MR imaging readily demonstrates gallbladder wall thickening and reveals Rokitansky-Aschoff sinuses as intraluminal lesions that are hyperintense on T2-weighted images, mimic a pearl necklace, and are characteristic findings for the diagnosis of adenomyomatosis of the gall bladder [9]. Although adenomyomatosis has generally been considered to carry no risk of malignant change, a few cases have reported carcinomas arising from adenomyomatosis in the literature [1–5]. In 1981 Nakafuji et
al. (4) reported that noninvasive carcinoma which had developed in the epithelium of a Rokitansky-Aschoff sinus. Kawarada et al. (3) described a case of noninvasive carcinoma occurring in an area of the segmental adenomyomatosis at the body and fundus of the gallbladder.

Until now, the relationship between the development of gallbladder cancer and adenomyomatosis has been unclear (7). Ootani et al. (7) studied the relation of adenomyomatosis to gallbladder carcinoma and the risk of developing gallbladder carcinoma in the presence of segmental adenomyomatosis of the gallbladder is higher than the risk in the general population. It is conceivable that the obstruction caused by annular stricture due to segmental adenomyomatosis leads to excessive intraluminal pressure and cholestasis in the fundal compartment and changes the epithelium of its mucosa. This changed mucosa is likely to produce gallbladder cancer.

In our case, MR imaging findings clearly showed segmental wall thickening of the gallbladder with Rokitansky-Aschoff sinuses as intramural small cystic lesions that were hyperintense on T2-weighted images. These findings suggested that gallbladder lesion was adenomyomatosis. However, it was difficult to determine at CT whether a gallbladder lesion was benign or malignant. In addition to segmental wall thickening of the gallbladder, newly developed, enlarged common hepatic and left paraaortic lymph nodes suggested that it might be metastases from gallbladder carcinoma or recurring gastric cancer. Therefore, we needed to further evaluated by FDG-PET/CT. The PET/CT and pathologic findings confirmed the gallbladder carcinoma with lymph node metastases.

In conclusion, although the malignant transformation of adenomyomatosis of the gallbladder is very rare, our case suggests that segmental adenomyomatosis itself may be a possible precursor for gallbladder carcinoma.

References

담낭의 분절형 선근종증에서 발생한 선암: 증례 보고

저자들은 담낭의 분절형 선근종증 자체에서 발생한 매우 드문 선암의 증례를 보고한다. CT 소견에서 담낭 벽의 국소비후와 국소립프절의 종대를 보여 악성종양을 배제하기 어려웠다. MR 영상과 PET/CT를 시행하였다. T2 강조 MRI에서 국소비후된 담낭 벽 내 선근종증을 시사하는 미세한 낭성 병변들이 발견되었으나, PET/CT에서는 병변 부위와 국소립프절에 양성반응을 보여 악성종양이 의심되었고, 병리소견에서 선근종증에서 발생한 선암으로 확진되었다.