

## Ciliated foregut cyst of the gallbladder: a case report and literature review

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Ciliated foregut cyst of gallbladder is a very rare benign cystic lesion. A 39-year-old woman was referred to our hospital after abdominal ultrasonography revealed a cystic lesion of gallbladder. On abdominal ultrasonography and computed tomography, a unilocular cystic lesion was found at right upper quadrant with attachment to the gallbladder neck. The gallbladder with cystic lesion was resected through laparoscopic cholecystectomy. The cystic lesion revealed a unilocular cyst with ciliated cuboidal or columnar epithelium and abundant goblet cells. Pathologic examination is essential to distinguish from other cystic lesions of the gallbladder and avoid unnecessary additional treatment. In the current case report, we presented the clinico-pathologic findings of the ciliated foregut cyst of the gallbladder and review of literature. ([Korean J Hepatobiliary Pancreat Surg 2016;20:85-88](#))

**Key Words:** Foregut; Gallbladder; Cysts; Congenital abnormalities

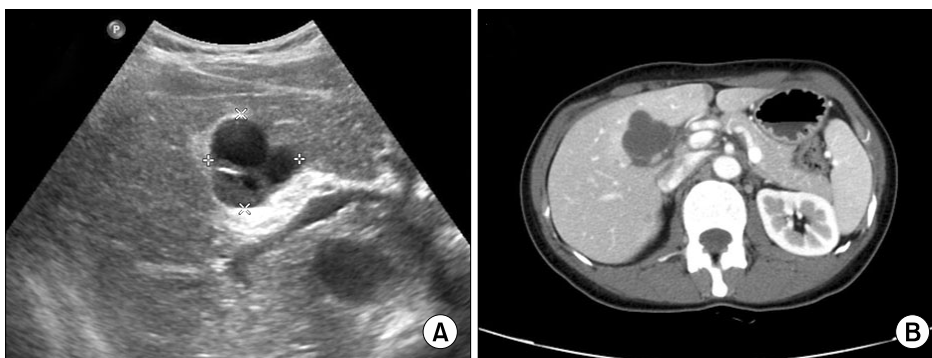
### INTRODUCTION

Cystic lesion of the gallbladder is uncommon and benign cystic lesion of the gallbladder is also very rare.<sup>1</sup> Foregut cyst is a rare disease and foregut cysts below the diaphragm are usually found in the liver.<sup>2</sup> Extrahepatic foregut cysts are very rare and can be found in the gallbladder, pancreas or upper gastrointestinal tract.<sup>3</sup> To our knowledge, only 8 cases of foregut cyst in the gallbladder were reported.<sup>1,2,4-9</sup> Herein, we presented a case of foregut cyst of the gallbladder and review of clinico-pathologic

findings reported in the literature.

### CASE

A 39-year-old female was referred to our hospital after abdominal ultrasonography revealed thickened wall of the gallbladder and a cystic lesion attached to the gallbladder. She had been admitted to a local hospital with right upper quadrant pain for 5 years. Tenderness of the right upper quadrant and Murphy's sign were present, but rebound tenderness was not present on physical examination. The



**Fig. 1.** Preoperative imaging studies. (A) Abdominal ultrasonography shows a cystic lesion with an amorphous debris level. (B) Abdominal computed tomography reveals a unilocular cystic lesion attached to the neck of the gallbladder.

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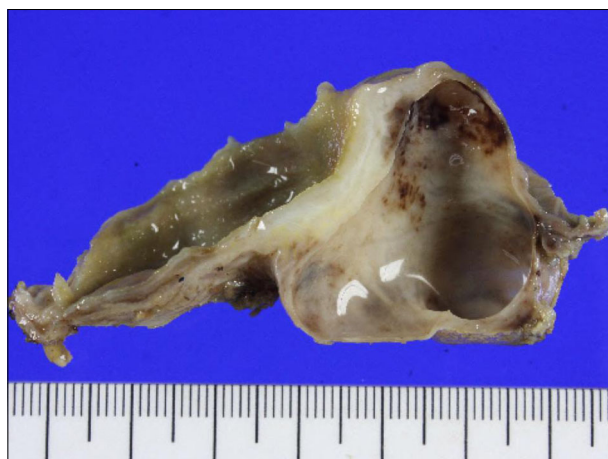
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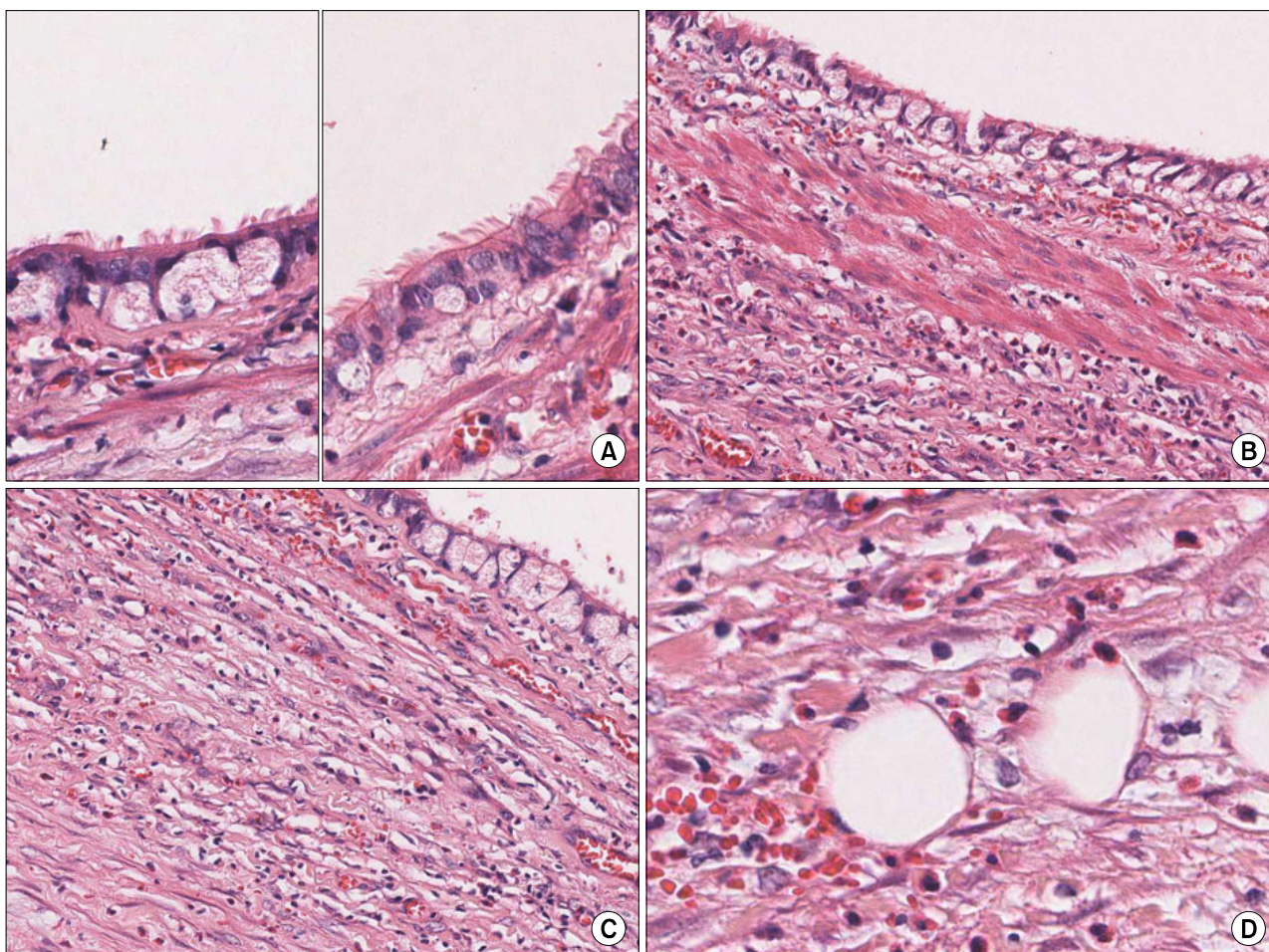
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results of laboratory examination indicated normal liver function tests, biliary enzymes and serum tumor markers. On abdominal ultrasonography and computed tomography, a unilocular cystic lesion was found at right upper quadrant with attachment to the gallbladder neck (Fig. 1A and 1B). Amorphous debris level was present in the cystic lesion. The gallbladder with cystic lesion was resected through laparoscopic cholecystectomy, in which there was no fibrosis or adhesion.

Grossly, the lesion revealed a unilocular cyst, measuring 3.5×3.2×3.0 cm, with mucin (Fig. 2). Microscopically, the cyst was lined with the ciliated cuboidal or columnar epithelium and abundant goblet cells (Fig. 3A). Squamous metaplasia or dysplasia was not present in the cystic epithelium. Dense smooth muscle layer or fibroelastic con-



**Fig. 2.** Gross photograph of the resected specimen. A mucinous unilocular cyst was attached to the neck of the gallbladder.



**Fig. 3.** Microphotographs of the resected specimen. (A) The cystic lining cells consist of the ciliated cuboidal (left) or columnar epithelium (right) and abundant goblet cells (H-E stain, ×400). (B) Dense smooth muscle layer is present in the subepithelial layer (H-E stain, ×200). (C) Subepithelial layer also contains the fibroelastic connective tissue (H-E stain, ×200). (D) Some eosinophils, lymphocytes and adipocytes are identified in the fibroelastic connective tissue (H-E stain, ×400).

nective tissue was identified under the lining epithelium (Fig. 3B and 3C). Some eosinophils, lymphocytes and adipocytes were present in the fibroelastic connective tissue of the subepithelial layer (Fig. 3D). These findings led us to the diagnosis of ciliated foregut cyst and chronic cholecystitis. The postoperative course was unremarkable and clinical examination showed no evidence of recurrence in the 20 months following surgery.

## DISCUSSION

Nam et al.<sup>8</sup> first introduced the term ‘ciliated foregut cyst of the gallbladder’ in their case report of 2000. Ciliated foregut cysts are very rare benign cysts, arising from the remnant of the embryonic foregut and usually located above the diaphragm, which present either as a bronchial or esophageal cyst.<sup>5,8</sup> When located below the diaphragm,

they are usually found in the liver, where they are known as ciliated hepatic foregut cysts. More than 60 cases of the ciliated hepatic foregut cysts have been reported, but extrahepatic cysts are extremely rare.<sup>5</sup> Only a few reports of the ciliated foregut cyst of the gallbladder were presented. Clinical manifestation and pathologic findings in literature and our case were summarized in Tables 1 and 2. Characteristically, all cases were middle-aged females and unilocular lesions. More than half of the cases were symptomatic, such as right upper or epigastric pain. Cystic lining cells usually consisted of pseudostratified columnar epithelium, and frequently single layer ciliated epithelium.<sup>1</sup> A few or many goblet cells could be observed in the lining epithelium.<sup>2,4,8</sup> Subepithelial layer consisted of fibroelastic tissue or thin smooth muscle layer similar to muscularis mucosa of gastrointestinal tract. Salivary gland acini could be found in the subepithelial area, similar

**Table 1.** Ciliated foregut cyst of the gallbladder with clinical manifestations in literature

Author	Year	Sex /Age	Location	Locularity	Size (cm)	Symptom	Duration of symptom	Underlying disease
Nam et al. <sup>8</sup>	2000	F/36	Fundus	Unilocular	1.5×1.0×1.0	No (incidental)	NA	Pulmonary tuberculosis
Hirono et al. <sup>6</sup>	2002	F/43	Collum	Unilocular	2.5	No (incidental)	NA	Acute renal failure
Muraoka et al. <sup>2</sup>	2003	F/37	Corpus	Unilocular	2.4×1.6×0.8	No (incidental)	NA	NA
Bulut and Karayalçın <sup>4</sup>	2010	F/41	Collum	Unilocular	3.5×2.0×1.5	RUQ pain	NA	NA
Tunçyürek et al. <sup>1</sup>	2013	F/42	Corpus	Unilocular	0.7×0.7	RUQ pain	NA	NA
Giakoustidis et al. <sup>5</sup>	2014	F/29	Neck	Unilocular	3.0	Epigastric pain	2 weeks	NA
Present case	2015	F/39	Neck	Unilocular	3.5×3.2×3.0	RUQ pain	5 years	None

NA, not applicable; RUQ, right upper quadrant

**Table 2.** Ciliated foregut cyst of the gallbladder with pathologic findings in literature

Author	Lining cells of the cyst	Subepithelial structure	Pathologic findings of the gallbladder
Nam et al. <sup>8</sup>	Pseudostatified ciliated columnar epithelium with few goblet cells	Fibroelastic tissue and smoothmuscle layer(s)	NA
Hirono et al. <sup>6</sup>	Pseudostatified ciliated columnar epithelium	No description	Adenomyomatosis
Muraoka et al. <sup>2</sup>	Pseudostatified ciliated columnar epithelium with interspersed mucin-producing columnarepithelium	Leiomyomuscular layer	Non-specific cholelithiasis
Bulut and Karayalçın <sup>4</sup>	Pseudostatified ciliated columnar epithelium with few goblet cells	Smooth muscle wall	Chronic calcareous cholecystitis with cholesterosis
Tunçyürek et al. <sup>1</sup>	Pseudostatified ciliated columnar epithelium and single layer of columnar epithelium	Smooth muscle fiber	NA
Giakoustidis et al. <sup>5</sup>	Pseudostatified ciliated columnar epithelium	Thin smooth muscle wall and Salivary gland acinus (small)	NA
Present case	Single layer ciliated cuboidal or columnar epithelium with many goblet cells	Fibroelastic tissue or smooth muscle layer	Chronic cholecystitis

NA, not applicable

to subepithelial glands of the bronchial wall.<sup>5</sup> Some eosinophils, lymphocytes or adipocytes were observed in the fibroelastic tissue. Histopathologic feature of the gallbladder was unremarkable.

Clinically, unilocular cystic lesion is a suspected benign lesion rather than malignant lesion; and all reported ciliated foregut cysts of the gallbladder were unilocular. Even though ciliated foregut cyst of the gallbladder is benign, excision is needed to resolve patient's symptom and to rule out other benign or malignant cystic lesions including biliary cystadenoma, cystadenocarcinoma, cystic gastric heterotopias, cystic duct cyst and cystic lymphangioma.<sup>10-14</sup> Histologic examination and accurate pathologic diagnosis are essential to avoid unnecessary additional treatment, such as chemotherapy or radiotherapy for malignant neoplasm. However, the ciliated foregut cyst of the gallbladder mostly suggest benign cystic lesion on radiology.

Although lining epithelium of our case consisted of the ciliated cuboidal or columnar cells without squamous metaplasia, ciliated foregut cyst frequently shows squamous metaplasia.<sup>15</sup> Squamous metaplasia is commonly observed in the uterine cervix or bronchial wall with chronic inflammation. Squamous metaplasia of epithelial cells can be induced in squamous cell carcinomas such as cervical squamous cell carcinoma of the uterus or squamous cell carcinoma of the bronchus.<sup>16,17</sup> There are a few reports of squamous cell carcinoma arising in the ciliated foregut cyst, however, there is no report on ciliated foregut cyst located in the gallbladder.<sup>18-20</sup> Ciliated foregut cyst requires surgical removal because of the frequent occurrence of squamous metaplasia and possibility of squamous cell carcinoma.

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## REFERENCES

1. Tunçyürek Ö, Nart D, Yaman B, Buyukcoban E. A ciliated foregut cyst in a gallbladder: the smallest recorded. *Jpn J Radiol* 2013;31:412-418.
2. Muraoka A, Watanabe N, Ikeda Y, Kokudo Y, Tatemoto A, Sone Y, et al. Ciliated foregut cyst of the gallbladder: report of a case. *Surg Today* 2003;33:718-721.
3. Sharma S, Dean AG, Corn A, Kohli V, Wright HI, Sebastian A, et al. Ciliated hepatic foregut cyst: an increasingly diagnosed condition. *Hepatobiliary Pancreat Dis Int* 2008;7:581-589.
4. Bulut AŞ, Karayalçın K. Ciliated foregut cyst of the gallbladder: report of a case and review of literature. *Patholog Res Int* 2010;2010:193535.
5. Giakoustidis A, Morrison D, Thillainayagam A, Stamp G, Mahadevan V, Mudan S. Ciliated foregut cyst of the gallbladder. A diagnostic challenge and management quandary. *J Gastrointest Liver Dis* 2014;23:207-210.
6. Hirono S, Tanimura H, Yokoyama S, Uchiyama K, Tani M, Onishi H, et al. Clinical features of ciliated foregut cyst of the gallbladder: a rare entity of cystic lesion in the gallbladder. *Dig Dis Sci* 2002;47:1817-1820.
7. Kakitsubata Y, Kakitsubata S, Marutsuka K, Watanabe K. Epithelial cyst of the gallbladder demonstrated by ultrasonography: case report. *Radiat Med* 1995;13:309-310.
8. Nam ES, Lee HI, Kim DH, Choi CS, Kim YB, Kim JS, et al. Ciliated foregut cyst of the gallbladder: a case report and review of the literature. *Pathol Int* 2000;50:427-430.
9. Benlolo D, Vilgrain V, Terris B, Zins M, Belghiti J, Menu Y. Imaging of ciliated hepatic or biliary cysts. 4 cases. *Gastroenterol Clin Biol* 1996;20:497-501.
10. Sistla SC, Sankar G, Basu D, Venkatesan B. Biliary cystadenocarcinoma of the gall bladder: a case report. *J Med Case Rep* 2009;3:75.
11. Rooney TB, Schofer JM, Stanley MD, Banks SL. Biliary cystadenoma of the gallbladder. *AJR Am J Roentgenol* 2005;185:1571-1572.
12. Özgün G, Adım SB, Uğraş N, Kiliçturgay S. A Mimicker of Gallbladder Carcinoma: Cystic Gastric Heterotopia with Intestinal Metaplasia. *Turk Patoloji Derg* 2014. doi: 10.5146/tjpath.2014.01227. [in press]
13. Yamamoto K, Yamamoto F, Maeda A, Igimi H, Yamamoto M, Yamaguchi R, et al. Tubulopapillary adenoma of the gallbladder accompanied by bile duct tumor thrombus. *World J Gastroenterol* 2014;20:8736-8739.
14. Shikano T, Takeda S, Sakai M, Sugimoto H, Kanazumi N, Nomoto S, et al. Cystic lymphangioma of the gallbladder: report of a case. *Surg Today* 2008;38:81-84.
15. Ben Mena N, Zalinski S, Svrcek M, Lewin M, Fléjou JF, Wendum D, et al. Ciliated hepatic foregut cyst with extensive squamous metaplasia: report of a case. *Virchows Arch* 2006;449:730-733.
16. Ishizumi T, McWilliams A, MacAulay C, Gazdar A, Lam S. Natural history of bronchial preinvasive lesions. *Cancer Metastasis Rev* 2010;29:5-14.
17. Miyatake T, Ueda Y, Yoshino K, Shroyer KR, Kanao H, Sun H, et al. Clonality analysis and human papillomavirus infection in squamous metaplasia and atypical immature metaplasia of uterine cervix: is atypical immature metaplasia a precursor to cervical intraepithelial neoplasia 3? *Int J Gynecol Pathol* 2007;26:180-187.
18. Zhang X, Wang Z, Dong Y. Squamous cell carcinoma arising in a ciliated hepatic foregut cyst: case report and literature review. *Pathol Res Pract* 2009;205:498-501.
19. Furlanetto A, Dei Tos AP. Squamous cell carcinoma arising in a ciliated hepatic foregut cyst. *Virchows Arch* 2002;441:296-298.
20. Vick DJ, Goodman ZD, Ishak KG. Squamous cell carcinoma arising in a ciliated hepatic foregut cyst. *Arch Pathol Lab Med* 1999;123:1115-1117.