

Intramural bronchogenic cysts of the esophagus and gastroesophageal junction: A case report

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Abstract. Bronchogenic cyst is a relatively rare congenital malformation that is often identified in the mediastinum. The occurrence of bronchogenic cysts in the intramural esophagus and gastroesophageal junction is rare. The present report describes three cases of intramural bronchogenic cysts of the esophagus and gastroesophageal junction and reviews the clinicopathological features of these lesions. A 35-year-old Japanese male (Case 1), a 50-year-old Japanese woman (Case 2) and a 34-year-old Japanese man (Case 3) presented with dysphagia, pharyngeal pain and heartburn, respectively. Upper endoscopic examination revealed submucosal tumors in the esophagus (Case 1 and 2) and gastroesophageal junction (Case 3). Subsequent endoscopic examination revealed perforation of the cyst into the surface of the esophageal mucosa (Case 2). Surgical resection was performed in all cases. Histopathological examinations revealed that the submucosal cysts were covered by respiratory-type ciliated epithelium without atypia. Cartilage and bronchial glands were not observed in any of the cases. The present review of the clinicopathological characteristics of bronchogenic cysts of the esophagus and gastroesophageal junction revealed that males and females were equally affected. The median age of the patients was 34.5 years with a wide age distribution. The most common main complaint was dysphagia. A pre-operative diagnosis of bronchogenic cyst is difficult because no specific imaging features are present. As surgical resection is recommended for this lesion, recognition of the clinicopathological features of bronchogenic cysts is important for an accurate pre-operative diagnosis.

Introduction

Bronchogenic cyst is a relatively rare congenital malformation that develops from abnormal budding of the ventral foregut during the early stage of gestation (1). Histologically, the cyst wall is covered by respiratory-type ciliated epithelium that may include cartilage and bronchial glands (2). The location of the cyst is dependent on the stage of embryogenesis at which budding of the foregut occurs (1). The most common location of bronchogenic cysts is the middle and superior mediastinum, following the lung parenchyma (1,2). Unusual locations of bronchogenic cysts include the thymus, pericardium, diaphragm, esophagus, stomach, and retroperitoneum (3-10).

Intramural esophageal bronchogenic cysts are rare, and only 25 cases have been reported in the English literature since 2000 (7,8,11-25). However, some cases of bronchogenic cysts in the mediastinum perforating into the esophagus have been described (26). Intramural bronchogenic cysts in the gastroesophageal junction are extremely rare (only 6 cases have been reported in the English literature) (27-32), and because of the rarity of intramural bronchogenic cysts in the esophagus and gastroesophageal junction, their unique clinicopathological features have not been well recognized. The treatment strategy for bronchogenic cyst is complete excision to avoid recurrence and rare malignant transformation (33,34). Therefore, accurate pre-operative diagnosis is very important for treatment.

In this study, we report three new cases of intramural bronchogenic cysts in the esophagus and gastroesophageal junction and review the clinicopathological characteristics of these rare lesions.

Case report

Case 1. A 35-year-old Japanese man presented with dysphagia. Upper endoscopic examination revealed an esophageal submucosal tumor. Pre-operative computed tomography (CT) was not available in our hospital. Subsequently, thoracoscopic enucleation of the tumor was performed. Histopathological examination indicated that a cyst (4.5x4x3.5 cm in diameter) was located in the muscularis propria and contained exudative fluid (Fig. 1). The cyst wall was covered by respiratory

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type ciliated epithelium without atypia (Fig. 1 inset). Neither cartilage nor bronchial glands were observed in the cyst wall. On the basis of these results, the patient was diagnosed with an intramural esophageal bronchogenic cyst. The post-operative course was uneventful, and the cyst did not reoccur by CT over 3 years of medical follow-up.

Case 2. A 50-year-old Japanese woman presented with pharyngeal pain and dysphagia. Upper endoscopic examination revealed a submucosal tumor in the esophagus, and no surface mucosal abnormality was noted. CT demonstrated a submucosal tumor in the esophagus (Fig. 2A). Subsequent endoscopic examination showed that the submucosal tumor was perforating into the esophageal lumen. Therefore, she underwent thoracoscopic subtotal esophagectomy. Histopathological examination revealed that the cyst (3.5x2 cm in diameter) was located in the muscularis propria of the esophagus, and perforated into the surface squamous mucosa of the esophagus, accompanied by lymphoplasmacytic infiltration around the cyst (Fig. 2B). The cyst wall was covered by respiratory-type ciliated epithelium without atypia, and a few goblet cells were occasionally observed (Fig. 2B, inset). Neither cartilage nor bronchial glands were observed in the cyst wall. On the basis of these clinical findings, the patient was diagnosed with an intramural bronchogenic cyst perforating into the esophageal. The post-operative course was uneventful, and the patient was free from recurrence by CT during the 3 months of medical follow-up.

Case 3. A 34-year-old Japanese man presented with heart-burn. Upper endoscopic examination demonstrated that a submucosal tumor (4.5x3.3 cm in diameter) was located immediately under the gastroesophageal junction without a remarkable change on the surface of the gastric mucosa. Pre-operative computed tomography (CT) was not available in our hospital. Subsequently, enucleation of the tumor was performed. Histopathological analysis showed that a large cyst was present under the mucosa of the gastroesophageal junction (Fig. 3A and B) and its wall was covered by respiratory-type ciliated epithelium without atypia (Fig. 3B, inset). Neither cartilage nor bronchial glands were observed in the cyst wall. Accordingly, the patient was diagnosed as intramural bronchogenic cyst in the gastroesophageal junction. The post-operative course was uneventful, and the patient was free from recurrence by CT for 1 year of medical follow-up.

Discussion

In this article, we describe the clinicopathological features of three cases of intramural bronchogenic cysts in the esophagus and gastroesophageal junction. Table I summarizes the clinicopathological features of intramural bronchogenic cysts of the esophagus and gastroesophageal junction that have been reported since 2000.

Among the 34 patients, men and women were equally affected. The median age of the patients was 34.5 years with a wide age distribution (from 3 days to 71 years) (Table I). The most common chief complaint was dysphagia with discomfort and pain, and no specific complaint for this lesion is present (Tables I and II). These symptoms may appear only when the

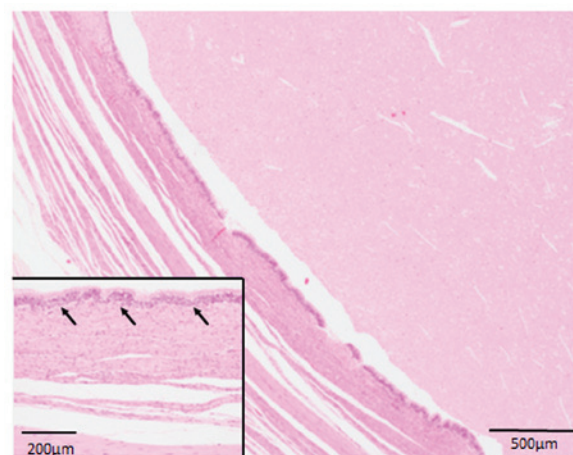


Figure 1. Histopathological features of the esophageal cyst (Case 1). The cyst was located in the muscularis propria and exudative fluid was present within the cyst. The cyst wall was covered by respiratory type ciliated epithelium without atypia (arrows; hematoxylin and eosin). Magnification, x100 and x200 (inset).

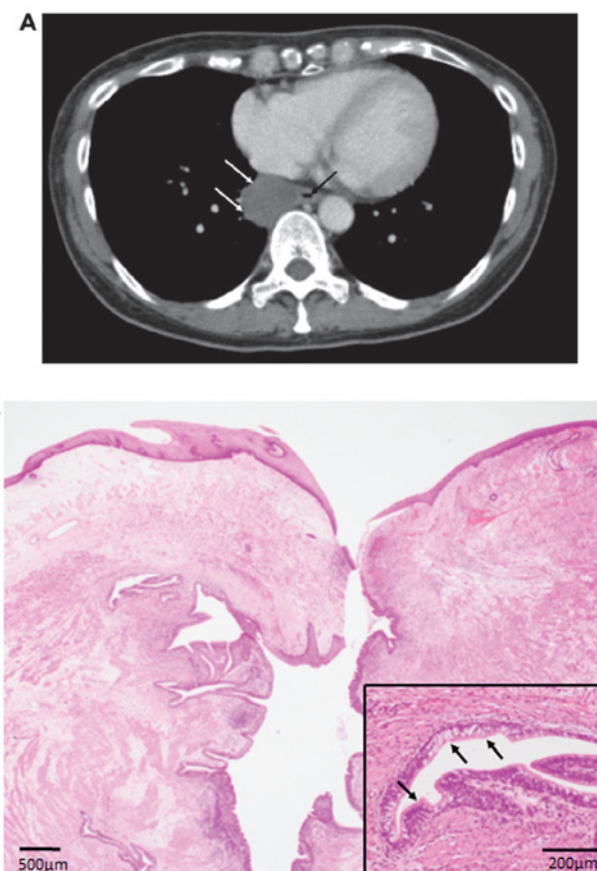


Figure 2. Computed tomography and histopathological features of the esophageal cyst (Case 2). (A) Well-circumscribed submucosal tumor of the esophagus (white arrows). Esophageal lumen was indicated by the black arrow. (B) The cyst was located in the muscularis propria of the esophagus, perforating into the surface squamous mucosa of the esophagus. The cyst was covered by respiratory-type ciliated epithelium without atypia, and goblet cells were occasionally observed (arrows, inset; hematoxylin and eosin stain). Magnification, x40 and x200 (inset).

cysts become larger, leading to compression of the esophagus and gastrointestinal junction. As most patients have small

Table I. Clinicopathological features of intramural bronchogenic cysts occurring in the esophagus and gastroesophageal junction.

Author, year	Case no.	Age	Sex	Size, cm	Chief complaint	Location	Procedure	Histological features	Outcome	(Refs.)
Cheng <i>et al.</i> , 2018	1	30 years	Male	8x7x4	Dysphagia, abdominal pain	Distal esophagus	Myotomy	Ciliated epithelium, cartilage, bronchial glands	FFR, 3 months	7
Lin <i>et al.</i> , 2017	2	20 months	Male	1.2x1x0.4	Recurrent vomiting	Distal esophagus	Laparoscopic resection	Ciliated epithelium, cartilage, bronchial glands	FFR, 7, months	11
Han <i>et al.</i> , 2016	3	31 years	Male	14.5x2.3	Chest pain, dysphagia	Para-esophagus	Thoracotomy	Ciliated epithelium	FFR, 3 months	12
Altieri <i>et al.</i> , 2015	4	40 years	Female	3	abdominal pain	Lower esophagus	Laparoscopic resection	Ciliated epithelium	FFR, 2 weeks	8
Suda <i>et al.</i> , 2015	5	3 days	Male	2x1	Inspiratory stridor	Cervical esophagus	Myotomy	Ciliated epithelium	FFR, 9 months	13
Tang <i>et al.</i> , 2014	6	23 years	Male	2.5x2	Chest discomfort, dyspnea	Distal esophagus	Endoscopic submucosal tunnel dissection	Ciliated epithelium, cartilage, bronchial glands	FFR	14
Vannucci <i>et al.</i> , 2013	7	39 years	Female	25	Dyspnea, palpable epigastric mass	Thoracoabdominal	Thoracotomy	NA	FFR, 36 months	15
Ghobakhlou <i>et al.</i> , 2012	8	23 years	Female	3x3	Dysphagia, abdominal pain	Distal esophagus	Thoracotomy	Ciliated epithelium	FFR	16
Wang <i>et al.</i> , 2012	9	56 years	Female	8x7x7	Chest pain, dysphagia	Lower paraesophagus	Thoracotomy	Ciliated epithelium, cartilage	FFR, 2 years	17
Barbetakis <i>et al.</i> , 2011	10	46 years	Male	NA	Dysphagia	Distal esophagus	VATS	NA	FFR	18
Chafik <i>et al.</i> , 2011	11	51 years	Male	3.6x3.1	Dysphagia, pain	Lower esophagus	Thoracotomy	Ciliated epithelium, bronchial glands	FFR	19
Turkyilmaz <i>et al.</i> , 2007	12	48 years	Male	3x2x1.5	Dysphagia	Distal esophagus	Thoracotomy	Ciliated epithelium, cartilage	FFR, 6 months	20
Akutsu <i>et al.</i> , 2006	13	26 years	Male	NA	Dysphagia	Lower esophagus	Thoracotomy	Ciliated epithelium, cartilage	FFR	21
Ko <i>et al.</i> , 2006	14	21 years	Male	4	Dysphagia, pain	Mid esophagus	VATS	Ciliated epithelium, cartilage (1/7 case)	FFR, 2 years	22
Ko <i>et al.</i> , 2006	15	31 years	Female	3.8	Dysphagia, pain	Mid esophagus	Thoracotomy		FFR, 6 years	22
Ko <i>et al.</i> , 2006	16	19 years	Female	3.2	Dysphagia, pain	Mid esophagus	Thoracotomy		FFR, 8 years	22
Ko <i>et al.</i> , 2006	17	20 years	Female	3	Dysphagia, chest discomfort	Mid esophagus	Thoracotomy		FFR, 4 years	22

Table I. Continued.

Author, year	Case no.	Age	Sex	Size, cm	Chief complaint	Location	Procedure	Histological features	Outcome	(Refs.)
A, Esophagus										
Ko <i>et al</i> , 2006	18	34 years	Female	3.9	Dysphagia, chest discomfort	Mid esophagus	Thoracotomy		FFR, 7 years	22
Ko <i>et al</i> , 2006	19	24 years	Female	3.6	Dysphagia	Lower esophagus	VATS		FFR, 1 year	22
Ko <i>et al</i> , 2006	20	60 years	Female	3.4	No symptom	Lower esophagus	Thoracotomy		FFR, 14 years	22
Westerterp <i>et al</i> , 2004	21	67 years	Male	6.6	Odynophagia	Thoracic esophagus	Esophagectomy	Ciliated epithelium	FFR	23
Westerterp <i>et al</i> , 2004	22	49 years	Female	3.1	Dysphagia	Mid esophagus	Endoscopic mucosal resection	Ciliated epithelium	FFR, 1 year	23
Westerterp <i>et al</i> , 2004	23	49 years	Female	3.3	No symptom	Esophagus	Local enucleation	Ciliated epithelium	FFR, 1 year	23
Hallani <i>et al</i> , 2004	24	64 years	Male	NA	Chest pain	Distal esophagus	Thoracotomy	NA	FFR	24
Sashiyama <i>et al</i> , 2002	25	34 years	Female	5	Dysphagia	Mid esophagus	Endoscopic mucosal resection	Ciliated epithelium	FFR	25
Present study	26	35 years	Male	4.5x4x3.5	Dysphagia	Lower esophagus	Enucleation	Ciliated epithelium	FFR, 3 years	-
Present study	27	50 years	Female	3.5x2	Pharyngeal pain and dysphagia	Mid esophagus	Thoracoscopic esophagectomy	Ciliated epithelium	FFR, 3 months	-
B, Gastroesophageal junction										
Tonouchi <i>et al</i> , 2016	1	32 years	Female	6	No symptom		Laparoscopic extirpation	Ciliated epithelium	FFR, 3 months	27
Kurokawa <i>et al</i> , 2013	2	71 years	Male	3	Throat discomfort		Laparoscopic resection	Ciliated epithelium	FFR, 1 year	28
Ballehaninna <i>et al</i> , 2013	3	40 years	Female	5x3.5	Dysphagia		Laparoscopic resection	Ciliated epithelium	FFR, 6 months	29
Fernández <i>et al</i> , 2011	4	33 years	Male	4.5x1.7	Epigastric and right upper pain		Laparoscopic resection	Ciliated epithelium	NA	30
Díaz Nieto <i>et al</i> , 2010	5	67 years	Male	6	Back pain		Laparoscopic resection	Ciliated epithelium	NA	31
Melo <i>et al</i> , 2005	6	39 years	Female	4x2.5x1	No symptom		Laparoscopic resection	Ciliated epithelium	FFR	32
Present study	7	34 years	Male	4.5x3.3	Heartburn		Enucleation	Ciliated epithelium	FFR, 1 year	-

FFR, free from recurrence; NA, not available; VATS, video-assisted thoracoscopic surgery.

Table II. Summary of the main characteristics of intramural bronchogenic cysts.

Symptoms	Patients, n	Histology	Patients, n	Procedure	Patients, n
Dysphagia	19	Ciliated epithelium	31	Thoracotomy	13
Pain	13	Cartilage	7	Laparoscopic resection	8
No symptoms	4	Bronchial glands	4	VATS	3
Chest discomfort	3			Endoscopic submucosal resection	3
Dyspnea	2			Myotomy	2

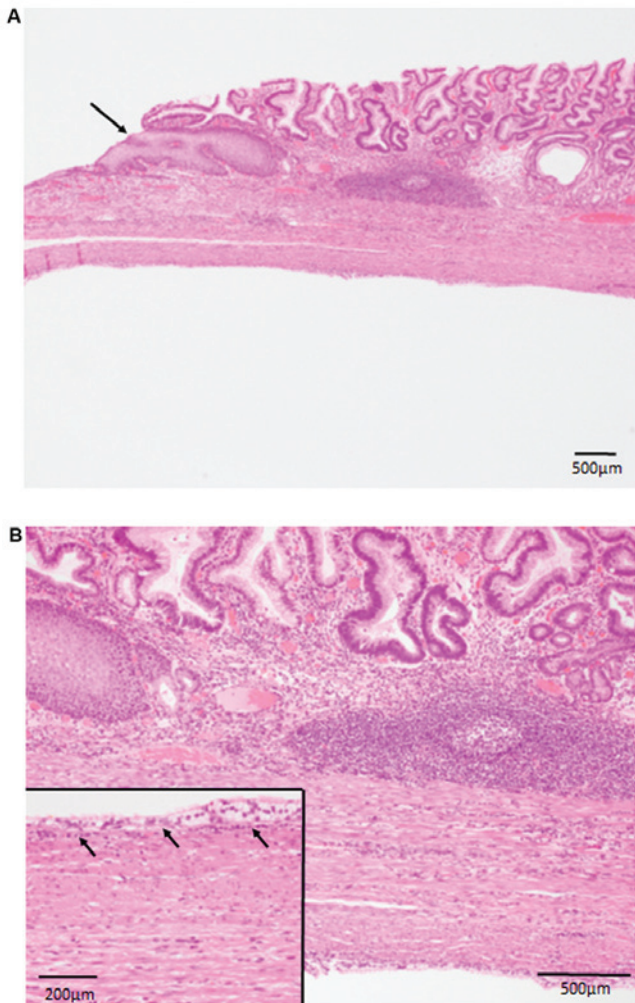


Figure 3. Histopathological features of the gastroesophageal cyst. (A) The cyst was located under the gastroesophageal mucosa [gastric type mucosa and squamous mucosa (arrow) were noted; H&E stain]. Magnification, x40. (B) The cyst wall was covered by respiratory-type ciliated epithelium without atypia (arrows; H&E stain). Magnification, x100 and x200 (inset). H&E, hematoxylin and eosin.

asymptomatic cysts (7), the accurate morbidity of these lesions is unclear. Histopathologically, the respiratory-type ciliated epithelium was observed in all cases, and cartilage and bronchial glands were occasionally found in the cyst wall.

Accurate pre-operative diagnosis of bronchogenic cyst is very important for its appropriate treatment. The diagnosis of these lesions is challenging because they do not have specific imaging characteristics. Ko *et al* (22) reported the imaging

characteristics of 7 cases of esophageal bronchogenic cysts. In the report, computed tomography (CT) revealed that well-defined thin-wall cystic lesions were present within the esophageal wall. Furthermore, varied cyst densities were observed (because of the content of the cyst) without enhancement after administration of a contrast agent, and no intracystic solid content or abnormal air was identified (22). However, it may be difficult to distinguish intramural bronchogenic cysts from mediastinal masses, including lymphadenopathy or mediastinal tumors, compressed against the esophageal wall (7,22). Furthermore, magnetic resonance imaging (MRI) showed variable signal intensities on T1-weighted images and a homogenous, high signal intensity on T2-weighted images (25).

Recently, endoscopic ultrasound (EUS) examination has been recognized as a useful tool for the diagnosis of bronchogenic cysts (7). EUS can identify whether the lesions of the esophagus and gastroesophageal junction are cystic or solid. Moreover, fine-needle aspiration (FNA) cytological examination using EUS can provide an even more accurate diagnosis because by this method it is possible to obtain a sample from the cyst wall. However, EUS-FNA may not be recommended for all patients with bronchogenic cysts because it can induce an infection, which would complicate the operation (7). The treatment strategy for bronchogenic cysts in the esophagus and gastroesophageal junction is complete resection (7). Therefore, a combination of the above-mentioned imaging techniques is required for an accurate pre-operative diagnosis of bronchogenic cyst.

The interesting finding of the present study is that one of bronchogenic cysts perforated into the esophageal lumen (Case 2). Only the second endoscopic examination detected the connection between the cyst and the surface mucosa of the esophagus. Therefore, secondary inflammation (probably due to infection) may have led to perforation of the cyst into the esophageal surface mucosa. Previous studies have already described an esophageal bronchogenic cyst with a connection to the surface squamous mucosa (17) and a mediastinal bronchogenic cyst perforated into the esophageal wall (26).

In conclusion, we reviewed the clinicopathological features of bronchogenic cysts in the esophagus and gastroesophageal junction. No specific symptoms or pre-operative imaging characteristics were present in this lesion, therefore, bronchogenic cyst must be added a list of differential diagnosis of the submucosal tumor of the esophagus and gastroesophageal junction. As surgical resection is recommended for this lesion, recognition of the clinicopathological features of bronchogenic cysts is important for accurate pre-operative diagnosis of this lesion.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

HM, MI and CM conceived and designed the present study. HM, MI, CM, TM, KI, MS and KT collected and analyzed data. HM and MI drafted the manuscript and figures. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The present study was conducted in accordance with the Declaration of Helsinki, and the study protocol was approved by the Institutional Review Board of Kansai Medical University Hospital (approval no. 2019050). Opt-out consent was obtained from each participant of this study.

Patient consent for publication

The need for informed consent was waived due to the retrospective design of the study, and opt-out consent was obtained from each participant of the present study.

Competing interests

The authors declare that they have no competing interests.

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