Intracardiac bronchogenic cyst is a rare congenital malformation that is a remnant from abnormal budding of the embryonic foregut. Among the intracardiac cases, bronchogenic cysts of the interatrial septum account for three-fourths of the reported cases. Because most of the intracardiac cysts are surgically removed promptly after detection, the natural course of intracardiac bronchogenic cyst is unknown. Here, we report the case of a growing interatrial bronchogenic cyst incidentally detected during the postoperative surveillance of gastric cancer.

Case Presentation
A 35-year-old male patient was referred to the cardiology department for an incidentally detected intracardiac mass. The patient had been diagnosed with advanced gastric cancer 14 months before referral, had undergone total gastrectomy, and was receiving adjuvant chemotherapy. The mass was first noted by his surveillance abdominal computed tomography (CT) scan, which covered his heart. Retrospective review of his preoperative stomach CT scan and follow-up abdomen CT scan revealed that the cyst had increased up to 6-fold in volume over a period of 13 months (Figure 1 and Figure I in the online-only Data Supplement).

At the time of referral, the patient was asymptomatic. Six years ago, he had an excision of a left second branchial cleft cyst. His medical history was otherwise unremarkable. The results of physical examination, chest radiograph, and routine blood sample analysis were normal. Twelve-lead ECG showed sinus rhythm with first-degree atrioventricular block (Figure IIA in the online-only Data Supplement). Transthoracic echocardiograms revealed a 2.1×2.1 cm cystic mass in the right atrial side of the interatrial septum. Contrast echocardiography with agitated saline revealed that the cyst had no flow communication with right atrium and ventricle (Figure 2, Movie I in the online-only Data Supplement). Other echocardiographic findings were normal. The estimated pressure of each atrium was 2 to 6 mm Hg for the right atrium and 4 to 12 mm Hg for the left atrium, respectively. ECG-gated CT angiography showed a cystic mass without contrast enhancement. Cardiac magnetic resonance image revealed high signal intensity on a fat-suppressed black blood T2-weighted image, low signal on black blood T1-weighted image, and no enhancement (Figure 2). From these findings, the cyst was assumed to have benign nature. However, because the mass was rapidly growing, based on CT scans of 5-month intervals, malignancy could not be completely excluded. Because the patient was undergoing his second-line adjuvant chemotherapy, the operation was deferred until the completion of chemotherapy.

Two months after his last chemotherapy cycle, the patient was admitted for an operation. Anterior right minithoracotomy and right atriotomy under cardiopulmonary bypass was successfully performed. An ≈3×2 cm round unilocular gray-white cyst was noted and completely excised. The cyst contained amber-colored mucous fluid. The histological analysis of the specimen revealed a benign cyst lined by pseudostratified ciliated cuboidal and columnar cells with focal cilia. Based on the pathological findings, the patient was diagnosed as having an interatrial bronchogenic cyst (Figure 3).

At immediate postoperation, 12-lead ECG showed complete atrioventricular block, which persisted 6 days after surgery. A dual-chamber pacemaker was then implanted before discharge (Figure IIB and IIC in the online-only Data Supplement). There was no other postoperative complication. At 2 months follow-up, the patient was symptom free without any evidence of cardiac tumor or gastric cancer recurrence.

Discussion
Here, we report a case of incidentally detected intracardiac bronchogenic cyst that was rapidly growing. Regarding the growth of an intracardiac bronchogenic cyst, there is only 1 case report of a 68-year-old woman who initially refused surgery at diagnosis. The interatrial bronchogenic cyst remained asymptomatic for 2 years without significant change in the size of the cyst. To our knowledge, this is the first report documenting the growth of an intracardiac bronchogenic cyst in an adult. In our case, the volume of cyst increased >6-fold in 13 months. The cyst was rather negligible for 34 years, but grew to a 3×2 cm cyst for 1 year. In general, the growth of bronchogenic cyst is thought to be the result of mucus accumulation, bleeding, infection, or malignant transformation.
Recent study of antenatally diagnosed bronchogenic cysts revealed that the cysts grow slowly in the first months of life but grow exponentially even in the absence of complication.1 The clinical presentation, image findings, and gross findings of the cyst in this case excluded hemorrhagic and infectious causes. Histopathologic examination of the cyst showed no evidence of malignant transformation. Therefore, we speculated that chronic inflammation precipitated by adjuvant chemotherapy could be the cause of cyst growth. The patient received oxaliplatin for 3 months and oral 5-fluorouracil prodrugs, capecitabine followed by S-1, for 1 year. The cyst grew 6-fold during that period. Mucositis and diarrhea are well-known adverse effects of 5-fluorouracil. Recent studies have revealed that the proinflammatory process and decreased reabsorption of water is the possible underlying mechanism of 5-fluorouracil–induced mucositis and diarrhea.2 Interestingly, inflammatory cell infiltration with multiple lymphocyte aggregates was found at the submucosal layer of the cyst (Figure III in the online-only Data Supplement). Chronic inflammation attributable to chemotherapy might have resulted in increased mucus accumulation and cyst growth.

With the development of imaging modalities, >50% of bronchogenic or benign cysts are diagnosed preoperatively. However, surgical removal of the lesion remains the mainstay of therapy. For intracardiac cysts, there is not a report of expectant management even in an asymptomatic patient. Although there is a chance that only surgically confirmed cases are reported in the literature, it is likely that the concern of malignant transformation and complication (ie, hemodynamic compromise, conduction disturbance, or embolism of cyst material) may urge surgical excision. The rare incidence and prompt surgical excision of intracardiac bronchogenic cysts limit the chance of observing serial growth of the benign lesion.

Acknowledgment

We thank Jin-Haeng Chung, MD, PhD and Yoon Jin Kwak, MD for their pathological review and discussion.

Disclosures

None.

References


Figure 1. Rapidly growing intracardiac mass incidentally detected during abdomen CT follow-up. Top left, At diagnosis of gastric cancer (13 months before referral). Top right, 7-month follow-up (6 months before referral). Bottom left, 12-month follow-up (1 month before referral). Bottom right, 13-month CT angiography (at referral). Scale bar, 1 cm. CT indicates computed tomography.
Figure 2. Multimodality characterization of interatrial cyst. **Top row**, 2.1×2.0 cm low echogenic cyst without blood flow communication on contrast echocardiography. **Middle row**, ECG-gated CT angiography of interatrial cyst shows absence of postcontrast enhancement. **Bottom row**: Left, Fat-suppressed black blood T2-weighted image; Middle, black blood T1-weighted image; Right, gadolinium-enhanced black blood T1-weighted image. CT indicates computed tomography.

Figure 3. Surgical resection of cyst and histopathologic diagnosis of bronchogenic cyst. Cyst lined by pseudostratified cuboidal and columnar cells with focal cilia (hematoxylin-eosin stain, ×400).
Intracardiac Bronchogenic Cyst: Report of a Growing Lesion
Jonghanne Park, Goo-Yeong Cho, Kay-Hyun Park and Il-Young Oh

Circulation. 2014;130:1107-1109
doi: 10.1161/CIRCULATIONAHA.114.010992
Circulation is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
Copyright © 2014 American Heart Association, Inc. All rights reserved.
Print ISSN: 0009-7322. Online ISSN: 1524-4539

The online version of this article, along with updated information and services, is located on the World Wide Web at:
http://circ.ahajournals.org/content/130/13/1107

Data Supplement (unedited) at:
http://circ.ahajournals.org/content/suppl/2014/10/15/130.13.1107.DC1

Permissions: Requests for permissions to reproduce figures, tables, or portions of articles originally published in Circulation can be obtained via RightsLink, a service of the Copyright Clearance Center, not the Editorial Office. Once the online version of the published article for which permission is being requested is located, click Request Permissions in the middle column of the Web page under Services. Further information about this process is available in the Permissions and Rights Question and Answer document.

Reprints: Information about reprints can be found online at:
http://www.lww.com/reprints

Subscriptions: Information about subscribing to Circulation is online at:
http://circ.ahajournals.org//subscriptions/
Supplemental Figure 1. Growth curve of an incidental intracardiac cyst at diagnosis of gastric cancer (13-month before referral), 7-month follow-up (6-month before referral) and 13-month (at referral). Volume of cyst was calculated with the ellipsoid assumption \( V = \frac{4}{3} \pi xyz \). From the CT image of each follow up, maximum radius of cyst on transverse plane \( x \), radius perpendicular to \( x \) \( y \) and maximum radius of cyst on coronary plane \( z \) was measured.
Supplemental Figure 2. Complete AV block after surgical resection of cyst leading to permanent DDDR PM insertion. A. Preoperative ECG, Sinus rhythm with 1st degree AV block, PR interval 310ms. Middle B. Postoperative ECG, complete AV block with PP interval 820ms and RR interval 1084ms. Bottom C. Paced rhythm with permanent PM.
Supplemental Figure 3. Micrographs of the intracardiac bronchogenic cyst. A. Low power field micrograph shows lymphoid aggregates (Arrows) in the submucosal layer (H&E stain, x40). B. High power field micrograph of a lymphoid aggregate shows marked infiltration of lymphocytes (H&E stain, x400).