

Thoracic Splenosis after Splenic and Diaphragmatic Injury

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Thoracic splenosis is a rare disease that develops as a result of autotransplantation of splenic tissue into the thoracic cavity following splenic and diaphragmatic injury. We report the case of a 53-year-old man with a chief complaint of heartburn and cough. He had a history of traumatic diaphragmatic rupture treated with surgical repair and splenectomy 15 years ago. Imaging studies revealed a paraesophageal mass, and surgical resection was performed considering the possibility of Castleman disease or an esophageal submucosal tumor. Pathologic results showed findings of normal splenic tissue. The patient was discharged on postoperative day 5 without any complications.

Key words: 1. Thoracic splenosis
2. Juxtapleural nodule
3. Traumatic diaphragm injury

Case report

A 53-year-old previously healthy man visited a local clinic due to a chronic heartburn sensation and cough. Based on abnormal findings on chest computed tomography (CT) and esophagogastroduodenoscopy, he was referred to Samsung Medical Center for further management.

Approximately 15 years ago, the patient had been hospitalized due to a traffic accident. The diagnosis was traumatic diaphragmatic and splenic rupture. He was successfully managed by bilateral chest tube drainage after surgical repair of the diaphragm and splenectomy via laparotomy. Two years previously, an incidental posterior mediastinal mass was detected by CT screening. However, he did not undergo any further work-up. At first, he received medical treatment, and took a proton pump inhibitor (such as dexlansoprazole), but it had no significant effect.

At our hospital, chest CT revealed a strongly-en-

hancing tubular soft-tissue lesion on the left side of the esophagus, measuring 5 cm longitudinally on the CT images (Fig. 1A). There was no other lesion in the abdomen. The radiologic diagnosis included a strongly-enhancing lymphatic lesion, as observed in Castleman disease (lymphoid hamartoma), or a submucosal tumor of the esophagus, as observed in leiomyoma or gastrointestinal stromal tumor (GIST). The endoscopic findings demonstrated an ovoid-shaped elevated lesion, consistent with a submucosal tumor (Fig. 1B). The lesion was located 35 cm from the incisor, without any mucosal involvement. Endoscopic ultrasonography also showed a mass originating from the esophageal muscle layer, with a maximum cross-sectional area of 29.1 mm×15.6 mm (Fig. 1C). Accordingly, the gastroenterologist consulted our department for possible surgical management.

We considered that the mass had contributed to his symptoms for the following reasons: (1) the mass was located just above the esophagogastric (EG)

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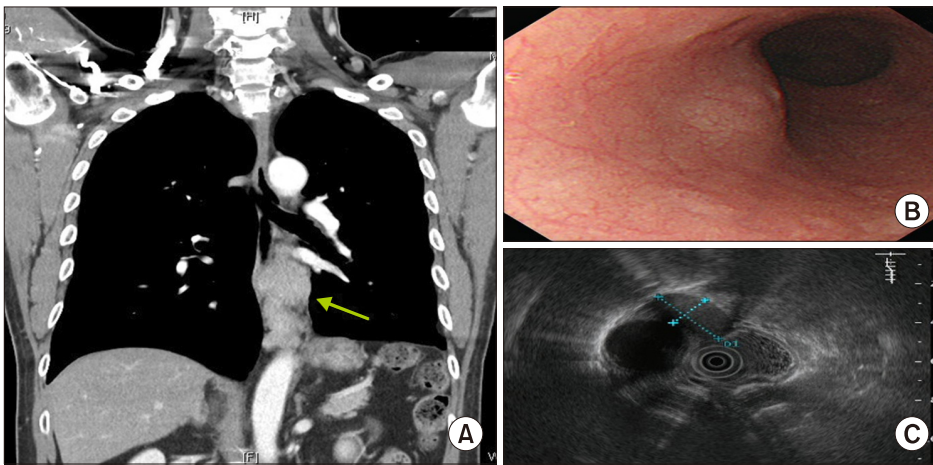


Fig. 1. (A) Chest contrast computed tomography showing a strongly-enhancing tubular soft-tissue lesion (arrow). (B) Esophagogastroduodenoscopy showing an ovoid elevated lesion. (C) Image from an endoscopic ultrasound scan.

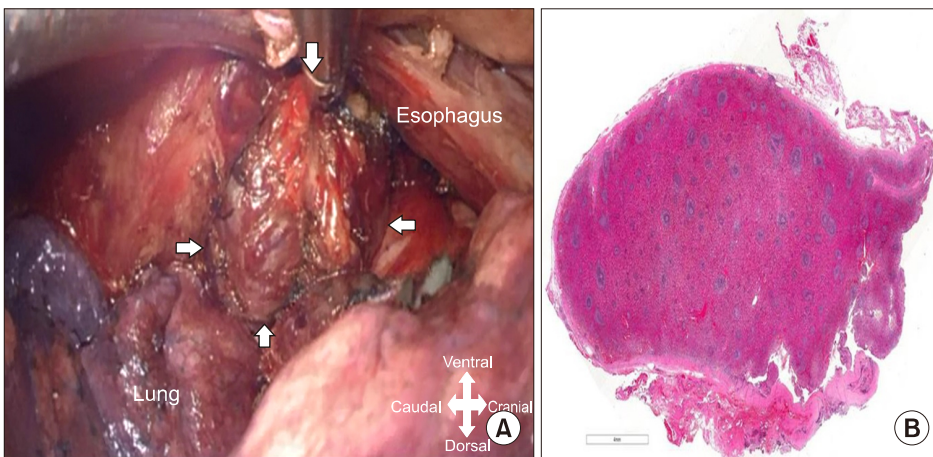


Fig. 2. (A) The mass adhered to the distal thoracic esophageal wall (arrows). (B) A thoracic mass showing normal splenic tissue (hematoxylin-eosin, Giemsa stain, and silver impregnation, $\times 40$).

junction and might have been related to functional impairment of the EG junction; and (2) CT and endoscopy did not demonstrate any lesions other than the mass. In addition, the mass had a considerable size (8–9 cm), so its mass effect may have affected the movement of the esophagus. Moreover, the location of the mass near the proximal stomach made it likely to cause symptoms and signs such as dysphagia, esophageal motility abnormalities, oropharyngeal dysfunction (including aspiration), and neuromuscular disorders, which are also associated with gastroesophageal reflux disease. Consequently, we planned elective mediastinal mass excision.

Surgery was performed through video-assisted thoracoscopic surgery on the right side with a 3-dimensional scope system. Prior to surgery, the mass was considered to be a submucosal tumor. The patient was put in a comfortable position, and a surgi-

cal approach from the right side was used, following the operator's general preference. Submucosal tumors can be adequately resected if they are approached from the right, even if the tumor is located primarily on the left. Although it was not easy to approach the patient from the right side, it was considered that this approach was preferable in light of the possibility of a future Ivor Lewis. During surgery, the mass was observed to originate from the pleural surface, rather than the esophagus. The size of the mass was 3 cm \times 5 cm \times 2 cm, and it adhered to the distal thoracic esophageal wall (Fig. 2A). After almost complete resection of the main mass, the specimen was sent to a pathologist for frozen section analysis. The result of the analysis revealed a lymphocyte and histiocyte-dominant tissue. Based on the results of the frozen biopsy, we decided not to perform extended dissection and finished the procedure. A chest tube

drain was inserted into the right pleural space.

After surgery, the patient underwent extubation in the operating room, and was transferred to the recovery room. The final pathological examination revealed thoracic splenosis. The histopathological features of the tissue were consistent with those of normal splenic tissue, characterized by the presence of red pulp and follicular structures with germinal centers (Fig. 2B). He recovered without any complications, was discharged on postoperative day 5, and his symptoms resolved after surgery.

Prior to publication, we informed the patient that his clinical data and perioperative imaging studies were included for case report, and the patient consent was obtained.

Discussion

Thoracic splenosis, which involves auto-transplantation of splenic tissue into the thoracic cavity, is a rare finding after traumatic rupture of the spleen [1]. Fewer than 40 cases have been described in the literature, and in all those cases, the patients had a combination of abdominal and thoracic trauma, and splenectomy was performed. Splenic tissues crossed the damaged diaphragm and spread to the serous surface of the pleural cavity. It is mostly asymptomatic and is diagnosed using regular chest radiography. The pulmonary parenchyma is an uncommon site of implantation [2]. Thoracic splenosis usually accompanies abdominal splenosis. All nodules are either pleural-based or in the interlobar fissures [1]. Splenic nodules can occur in the lung parenchyma [3].

Thoracic splenosis should be suspected in patients with juxtapleural nodules when there is a history of splenic and diaphragmatic damage. If there is a history of trauma, and if tomographic images are available, nuclear medicine can confirm thoracic splenosis without biopsy. Images from nuclear medicine are commonly obtained by scintigraphy using Tc-99m sulfur colloid, which enhances reticuloendothelial system cells as well as sulfur colloid [4]. In our patient, the initial radiological imaging was suggestive of Castleman disease. The endoscopic findings were suggestive of a submucosal tumor of the esophagus, such as leiomyoma or GIST. Pleural-based masses of thoracic splenosis are often misdiagnosed as neo-

plastic lesions. Surgery is indicated only in symptomatic patients and in those whose diagnosis is doubtful; in this case, it was not possible to exclude malignancy [5]. Resection of ectopic splenic tissue should be avoided not only because the surgical procedure itself is risky, but also for protection against post-splenectomy sepsis [6]. Leemans et al. [6] demonstrated that spleen transplants provided better pneumococcal blood clearance and increased immunoglobulin M levels and opsonization activity. Therefore, surgical removal should not be performed unless it is absolutely necessary, particularly in asplenic patients with hematologic diseases, in whom the ectopic splenic tissues can protect against bacterial infections.

Some reports of thoracic splenosis in other countries have been published, and several cases of thoracic splenosis have also been reported in Korea. However, those cases had been misdiagnosed as pleural disease or lung disease. To the best of our knowledge, this is the first case to be reported in Korea in which thoracic splenosis resembled an esophageal tumor or submucosal tumor. In addition, this case is valuable because this is the first case of thoracic splenosis presenting in the paraesophageal area. Theoretically, ectopic splenic tissue can be disseminated elsewhere around the pleura based on its known mechanism of spread. Most of the reported cases showed intrathoracic masses with variable locations, such as single or multiple pleural-based masses [1], diaphragmatic masses [2], and hilar masses [2]. Until now, however, there has been no reported case of thoracic splenosis mimicking a paraesophageal mass.

Although the need for surgical removal may vary depending on the location of thoracic splenosis, we believe that most cases of thoracic splenosis do not require surgical management. In our case, thoracic splenosis was found just beside the esophageal wall of the posterior mediastinum, which is unusual. Our decision was made based on the possibility of a mass-symptom relationship; however, thoracic splenosis in most other thoracic locations does not cause digestive or respiratory symptoms. There is no reason to remove an asymptomatic mass that does not have malignant potential. With the lessons learned from our unique experience, we suggest that it is necessary to carefully determine whether surgery

should be performed in such cases.

In conclusion, thoracic splenosis is a rare condition, but should be considered in the differential diagnosis of juxtapleural nodules, especially when there is a history of thoracoabdominal trauma. To avoid unnecessary surgery or an invasive diagnostic approach, suspicion for thoracic splenosis is of paramount importance. The appropriate use of commonly available diagnostic tools, such as tomographic and scintigraphic images, can facilitate the accurate diagnosis of thoracic splenosis. More case reports and further clinical research are necessary to establish the efficacy of the non-invasive diagnosis of thoracic splenosis.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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