http://dx.doi.org/10.4046/trd.2013.74.3.134
ISSN: 1738-3536(Print)/2005-6184(Online)
Tuberc Respir Dis 2013;74:134-139

Case Report

Combined Intrathoracic and Intraperitoneal Splenosis after Splenic Injury: Case Report and Review of the Literature

Chansoo Moon, M.D.[1], Yun-Jung Choi, M.D.[2], Eun Young Kim, M.D.[1], In Sun Lee, M.D.[1], Sae Byol Kim, M.D.[1], Sung Mo Jung, M.D.[1], Se Kyu Kim, M.D.[1], Joon Chang, M.D.[1], Ji Ye Jung, M.D.[1]

[1]Division of Pulmonology, Department of Internal Medicine, Institute of Chest Disease, [2]Department of Nuclear Medicine, Yonsei University College of Medicine, Seoul, Korea

Splenosis is defined as an autotransplantation of the splenic tissue after splenic rupture or splenectomy, and occurs most frequently in the peritoneal cavity. Splenosis is usually asymptomatic and is found incidentally. We report a case of combined intrathoracic and intraperitoneal splenosis in a 54-year-old male who worked as a miner for 10 years in his twenties, and was a current smoker. He was referred to our hospital for further evaluation of an incidental left diaphragmatic mass. Positron emission tomography-computed tomography and bronchoscopy were performed to evaluate the possibility of malignancy. There was no evidence of malignancy, but the spleen was not visualized. Reviewing his medical history revealed previous splenectomy, following a dynamite explosion injury. Therefore, splenosis was suspected and technetium-99m-labeled heat-damaged red blood cell scan confirmed the diagnosis. Radionuclide imaging is a useful diagnostic tool for splenosis, which could avoid unnecessary invasive procedures.

Key Words: Splenosis; Splenectomy; Radionuclide Imaging

Introduction

Splenosis is a rare condition that results from autotransplantation of splenic tissue into abnormal sites, such as the peritoneal cavity, thoracic cavity, subcutaneous tissue, and pericardium. This ectopic spleen tissue usually occurs after traumatic rupture of the spleen and is generally asymptomatic. However, the ectopic spleen tissue may occasionally cause pain or gastrointestinal obstruction, or mimic primary and metastatic malignancy. Therefore, splenosis is usually found incidentally on imaging studies, and is rarely diagnosed without invasive biopsy including surgical methods. However, radionuclide scanning can provide an alternative to these invasive procedures.

Previously reported cases of splenosis in South Korea involved single compartments of the body, either intraperitoneal or intrathoracic[1-9]. Moreover, most were diagnosed through surgical pathological confirmation. We describe a 54-year-old Korean male who presented with intrathoracic and intraperitoneal splenosis diagnosed by radionuclide imaging.

Address for correspondence: **Ji Ye Jung, M.D.**
Division of Pulmonology, Department of Internal Medicine, Yonsei University College of Medicine, 50 Yonsei-ro, Seodaemun-gu, Seoul 120-749, Korea
Phone: 82-2-2228-1980, Fax: 82-2-393-6884
E-mail: STOPYES@yuhs.ac
Received: May 2, 2012
Revised: May 23, 2012
Accepted: Jul. 25, 2012

Case Report

A 54-year-old male was referred to Severance Hospital for evaluation of incidental left diaphragmatic mass and right upper lobe consolidation on chest X-ray (Figure 1A). Pleural core needle biopsy done at the previous hospital showed acute and chronic inflammation with mixed inflammatory cell infiltration. He had no unusual medical conditions and worked as a miner for 10 years in his twenties. He had smoked half a pack of cigarettes

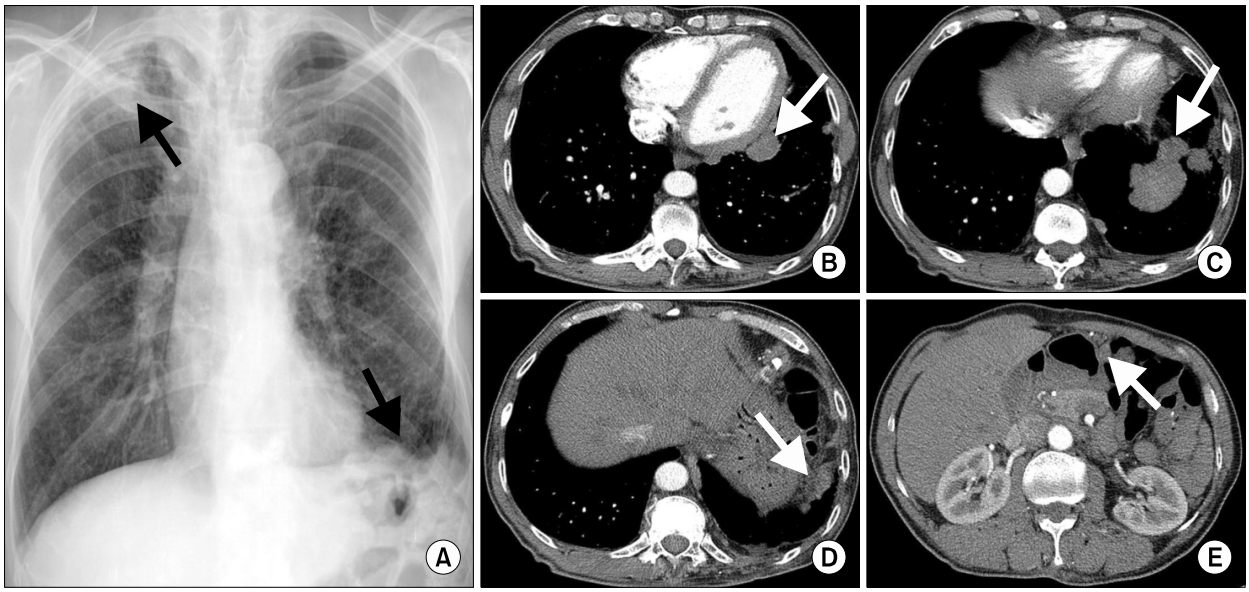

Figure 1. (A) Chest X-ray showing a left diaphragmatic mass with irregular margin and consolidation in the right upper lobe. (B–E) Chest computed tomography. Multiple lobulated masses and nodules present along the left lower mediastinum (B), left diaphragm (C, D), and omentum (E).

per day for 40 years, and quit 3 months before.

On admission, he did not have any symptoms, physical examination was unremarkable, and laboratory tests were normal. Chest computed tomography (CT) revealed multiple lobulated masses and nodules along diaphragm, lower mediastinum, and costal pleura of left side. Moreover, multiple calcified granulomas with fibrosis were seen in the right upper lobe (Figure 1B–E). Considering the previous history of smoking and mining, malignant tumors such as mesothelioma or pleural metastasis had to be differentiated and we proceeded ^{18}F-fluoro-2-deoxygluocose (^{18}F-FDG) positron emission tomography-CT (PET-CT). FDG PET-CT showed previously detected multiple masses and nodules along left diaphragm and multiple small nodular lesions in the left perigastric area and omentum with faint FDG uptake. Acute and chronic inflammation with fibrosis and calcified granulomas were observed in right upper lobe with intense FDG uptake (Figure 2A). However, his spleen was not visualized on PET-CT and review of chest CT. From further medical history obtained, he had undergone splenectomy following splenic trauma from a dynamite explosion while working in the mine 30 years ago.

With this history, splenosis was suspected, and a technetium (Tc)-99m-labeled heat-damaged red blood cell (RBC) scan was performed. The RBC scan showed multiple foci of increased uptake in the left thorax and left upper quadrant of the abdomen, consistent with thoracic and abdominal splenosis (Figure 2B). This patient has been followed up at our outpatient clinic without further invasive evaluation or treatment for the splenosis. He was diagnosed with pulmonary tuberculosis based on positive sputum culture and is on anti-tuberculous medications.

Discussion

Ectopic splenic tissue usually manifests in the body in two forms, accessory spleens and splenosis. Accessory spleens are congenital and found in approximately 20% of the population. They result from failure of spleen tissue union during embryogenesis and arise from the left side of the dorsal mesogastrium. Accessory spleen usually has normal splenic histology with its blood supply from a branch of the splenic artery. On

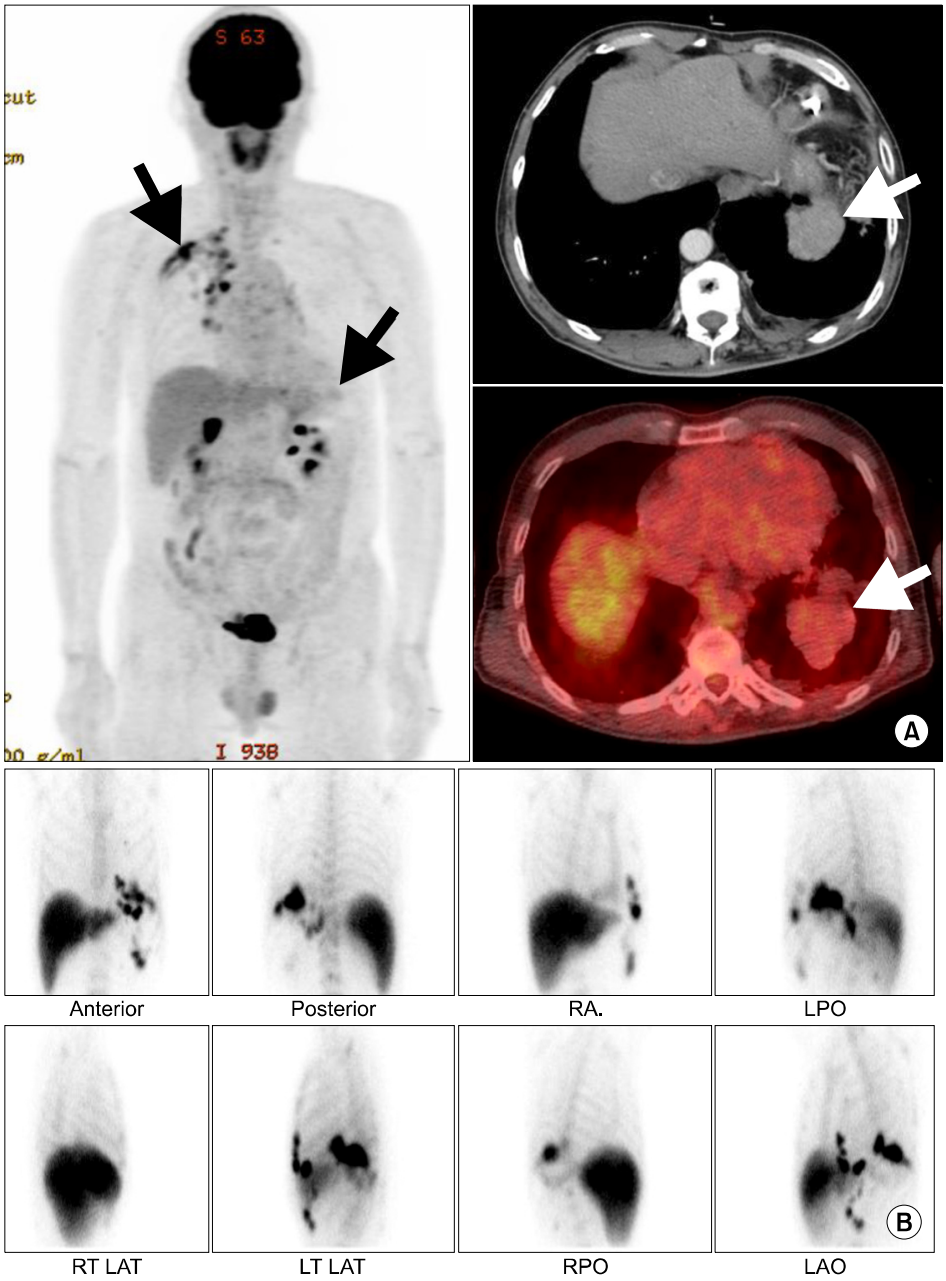

Figure 2. (A) Positron emission tomography computed tomography (PET-CT). Multiple masses and nodules along left diaphragm and multiple small nodular lesions in the left perigastric area and omentum show faint fluoro-2-deoxygluocose (FDG) uptake. Moreover, acute and chronic inflammation with fibrosis and calcified granulomas were observed in the right upper lobe with intense FDG uptake. (B) Technetium-99m-labeled heat-damaged red blood cell scan. Multiple foci show increased uptake in the left thorax and left upper quadrant of the abdomen, consistent with thoracic and abdominal splenosis. RAO: right anterior oblique; LPO: left posterior oblique; RT LAT: right lateral; LT LAT: left lateral; RPO: right posterior oblique; LAO: left anterior oblique.

the other hand, splenosis is an acquired condition, which is rare compared to accessory spleen. It is autotransplantation of viable splenic tissue to any anatomic compartments of the body and usually exhibits distorted architecture.

Von Kuttner[10] first reported splenosis in the peritoneal cavity after splenectomy during autopsy in 1910. The first intrathoracic splenosis was reported by Shaw and Shafi[11]. There are two possible pathogeneses of splenosis. One is through mechanical implantation of splenic tissue after splenectomy or splenic rupture. It is assumed that the damaged splenic pulp is released into the adjacent cavity, which begins the seeding process. The other mechanism is hematogenous spread of splenic pulp resulting in intrahepatic and cerebral splenosis[12]. Therefore, splenosis can occur in any part of the body, but is most frequently observed in the peritoneal cavity. Among the extraperitoneal splenosis, thoracic splenosis

Table 1. Clinical characteristics of patients with splenosis reported in Korea

Reference	Classification	Location	Age (yr)/Sex	Cause of splenectomy	Interval (yr)	Symptoms	Size (cm)/ No. of lesions	Primary suspected disease	Diagnostic method
Hoh et al.[1]	Pelvic	Peritoneum	34/F	Fall	21	Cyclic abdominal pain, dyspareunia	8.5/single	Ovarian dermoid cyst	Laparotomy
Kim et al.[2]	Abdomen	Liver	43/M	Traffic accident	20	Incidentally found hepatic mass	3.5/single	Hepatocellular carcinoma	Liver segmentectomy
Ryu et al.[3]	Abdomen	Omentum	56/M	Fall	37	Epigastric pain	1.5/multiple	Peritoneal carcinomatosis	Laparotomy
Yoon et al.[4]	Abdomen	Peritoneum	44/M	Traffic accident	9	Epigastric pain	3.3/multiple	Splenosis	Tc-99m-labeled heat-damaged RBC scan
Choi et al.[5]	Abdomen	Liver, omentum	32/M	Traffic accident	26	Further treatment after TACE for HCC	3/multiple	Hepatocellular carcinoma	Laparotomy liver segmentectomy
Bang et al.[6]	Abdomen	Peritoneum	43/M	Traffic accident	15	Incidentally found abdominal mass	4/multiple	Splenosis	Tc-99m-labeled heat-damaged RBC scan
Chung et al.[7]	Abdomen	Omentum, peritoneum	45/F	Traffic accident	25	Further treatment for colon polyps	1/multiple	Peritoneal carcinomatosis	Laparotomy
Kim et al.[8]	Thoracic	Left diaphragmatic pleura	42/M	Penetration	20	Incidentally found pleural mass	4.8/single	Sclerosing hemangioma	Video-assisted thoracoscopic surgery
Kang et al.[9]	Abdomen	Liver, omentum	54/M	Traffic accident	15	Further treatment for gastric cancer	2.3/multiple	Liver metastasis from gastric cancer	Liver segmentectomy
Present case	Thoracic, abdomen	Left diaphragmatic pleura, omentum	53/M	Dynamite explosion	30	Incidentally found pleural mass	5.1/multiple	Mesothelioma	Tc-99m-labeled heat-damaged RBC scan

F: female; M: male; Tc: technetium; RBC: red blood cell; TACE: transcatheter arterial chemoembolization; HCC: hepatocellular carcinoma.

is the most common form and few cases have been reported in the pericardium, subcutaneous tissue, and occipital pole of the brain[13].

The reason thoracic splenosis is most common among the extraperitoneal sites is probably due to the anatomic boundaries of the abdominal and thoracic cavities. Most of the patients with thoracic splenosis had history of thoracoabdominal trauma including diaphragm injury and splenic rupture[13]. This accounts for all the thoracic splenosis observed being on the left side. Thoracic splenosis is usually asymptomatic and results in no significant morbidity or mortality. However, one case of thoracic splenosis reported several milliliters of hemoptysis[14]. The differential diagnosis for thoracic splenosis includes pleural metastasis or malignant mesothelioma.

Previously, many cases of thoracic splenosis were diagnosed through percutaneous biopsy or intraoperative resection of masses when the splenosis was not considered at first. However, fine-needle aspiration cytology of subpleural splenosis can lead to misdiagnosis if the cytologist is not informed of the patient's history of splenic injury. Moreover, pleural adhesions in patients with previous surgery make thoracotomy surgical biopsy difficult. Therefore, preoperative diagnosis of splenosis by radionuclide imaging study can avoid unnecessary biopsy or surgical procedures. Tc-99m sulfur colloid is sequestered in the reticuloendothelial system of ectopic splenic tissue and therefore can be used to detect it. Indium-111-labeled platelets and Tc-99m-labeled heat-damaged RBC are very sensitive and specific for splenic sequestration and phagocytosis[15].

In Korea, nine cases of splenosis were previously reported. Seven cases were located in the abdominal cavity, one case in the pelvis, and one in the thoracic cavity (Table 1). Seven patients were diagnosed through surgical pathological confirmation and only two patients were diagnosed through Tc-99m-labeled heat-damaged RBC scan.

In conclusion, splenosis can develop in multiple sites depending on the extent of injury. Therefore, thoracic splenosis should be considered in asymptomatic patients with multiple left-side pleural masses and previous thoracoabdominal trauma and splenic injury. Tc-99m-labeled heat-damaged RBC scan is the diagnostic method of choice for noninvasive evaluation of splenosis instead of invasive pathological confirmation.

References

1. Hoh JK, Kim KT, Cho SH, Hwang YY. A case of pelvic splenosis after splenectomy: a cause of pelvic pain. Korean J Obstet Gynecol 2005;48:3004-8.
2. Kim KA, Park CM, Kim CH, Choi SY, Park SW, Kang EY, et al. An interesting hepatic mass: splenosis mimicking a hepatocellular carcinoma (2003:9b). Eur Radiol 2003;13:2713-5.
3. Ryu SW, Kim IH, Sohn SS. Splenosis mimicking carcinomatosis peritonei in advanced gastric cancer. J Korean Surg Soc 2005;68:61-4.
4. Yoon M, Hwang KH, Choe W. Multifocal peritoneal splenosis in Tc-99m-labeled heat-denatured red blood cell scintigraphy. Nucl Med Mol Imaging 2006;40:190-1.
5. Choi GH, Ju MK, Kim JY, Kang CM, Kim KS, Choi JS, et al. Hepatic splenosis preoperatively diagnosed as hepatocellular carcinoma in a patient with chronic hepatitis B: a case report. J Korean Med Sci 2008;23:336-41.
6. Bang SY, Paik CH, Chung YW, Chang MH, Kim JE, Kim YS, et al. A case of intraperitoneal splenosis: clinical history and imaging features avoided the need for invasive diagnostic procedures. Korean J Med 2008;74 (Suppl 1):S33-6.
7. Chung SJ, Jahng JH, Mun HS, Park YA, Jung WH, Yu JS, et al. Case of colonic polyps with abdominal splenosis mimicking carcinomatosis. Korean J Med 2008; 75:574-6.
8. Kim K, Choi HJ, Kim YM, Kwon WJ, Lee WC, Suh JH. Thoracic splenosis: a case report and the importance of clinical history. J Korean Med Sci 2010;25:299-303.
9. Kang KC, Cho GS, Chung GA, Kang GH, Kim YJ, Lee MS, et al. Intrahepatic splenosis mimicking liver metastasis in a patient with gastric cancer. J Gastric Cancer 2011;11:64-8.
10. von Kuttner H. In diskussion, milzextirpation und rontgenbehandlung bei leukamie. Berl Klin Wochenschr 1910;47:1520.
11. Shaw AF, Shafi A. Traumatic autoplastic transplantation of splenic tissue in man with observations on the late results of splenectomy in six cases. J Pathol Bacteriol 1937;45:215-35.
12. Kwok CM, Chen YT, Lin HT, Su CH, Liu YS, Chiu YC.

Portal vein entrance of splenic erythrocytic progenitor cells and local hypoxia of liver, two events cause intrahepatic splenosis. Med Hypotheses 2006;67:1330-2.
13. Fremont RD, Rice TW. Splenosis: a review. South Med J 2007;100:589-93.
14. Cordier JF, Gamondes JP, Marx P, Heinen I, Loire R. Thoracic splenosis presenting with hemoptysis. Chest 1992;102:626-7.
15. Armas RR. Clinical studies with spleen-specific radiolabeled agents. Semin Nucl Med 1985;15:260-75.