

Xanthomatous Pleuritis Associated With Pancreaticopleural Fistula

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Xanthomatous pleuritis is a very rare type of chronic inflammation, with only 3 reports to date.^{1–3} However, the exact etiology of this disease is unknown. Herein, we present a case of xanthomatous pleuritis that might have been associated with pancreatic pleural effusion.

CASE REPORT

A 62-year-old male who regularly consumed alcohol (<20 g/day), was a non-smoker, and had no medical history was referred to our hospital for exertional dyspnea. Laboratory examination revealed leukocytosis (11,200/ μ L); however, all other findings were normal (C-reactive protein, 0.62 mg/dL; serum amylase, 171 IU/L; serum lipase, 30 IU/L; tumor markers, negative; interferon-gamma release assay, negative). The chest radiograph showed left pleural effusion. Moreover, chest computed tomography (CT) revealed left pleural effusion. We performed thoracoscopy under local anesthesia for drainage and pathologic diagnosis as pleural effusion because of malignancy was considered. Pleural fluid examination revealed bloody exudative (protein, 5.0 g/dL; lactate dehydrogenase, 1264 IU/L; Rivalta reaction, positive; serum total protein, 6.9 g/dL; serum, lactate dehydrogenase 212 IU/L) and high level of amylase (13,355 IU/L). Thoracoscopy of the left thorax revealed diffuse yellow plaque-like deposits in the pleura (Fig. 1). Histology of the left pleural biopsy specimen showed inflammation and foamy cells (Fig. 2). There was no evidence of malignancies and infections, including *Mycobacterium*. Therefore, we considered the presence of pancreaticopleural fistula and performed further abdominal examinations. An enhanced CT and endoscopic retrograde cholangiopancreatography revealed pancreatic stones, main pancreatic duct dilatation, meandering pancreatic duct, and left pancreaticopleural fistula (Fig. 3). Distal pancreatectomy was performed after thoracic drainage with a chest tube to treat the pancreatic pleural fistula for prevention of pleural effusion recurrence and rule out pancreatic cancer. The surgically resected specimen of the pancreas showed chronic pancreatitis with pancreatic stones and no findings of malignancies. On the basis of these results, xanthomatous pleuritis with pancreaticopleural fistula because of chronic pancreatitis

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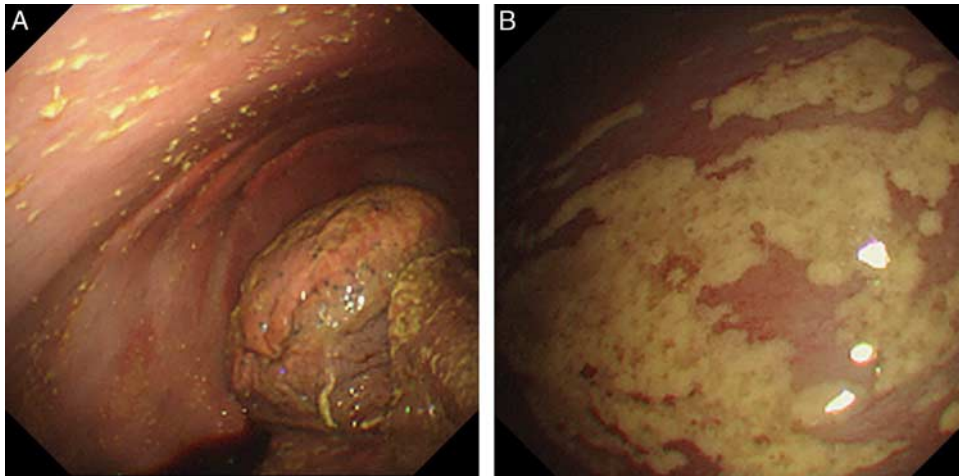


FIGURE 1. A and B, Thoracoscopic image shows diffuse yellow plaque-like deposits in the pleura of the left thorax.

was finally diagnosed. The patient has had an uneventful course without recurrence for more than 4 years.

DISCUSSION

To the best of our knowledge, this is the first report of xanthomatous pleuritis with pancreatic pleural effusion. Xanthomatous pleuritis is a very rare benign type of chronic inflammation, and only 3 cases have been reported in the literature.¹⁻³ Although the exact etiology is uncertain, previous reports^{2,4} suggested that chronic inflammation, defective lipid transport, and hemorrhage might be the causes of xanthomatous inflammation. In the present case, chronic inflammation because of pancreaticopleural fistula might have caused xanthomatous pleuritis. Furthermore, aberrance of the lipid component of the pancreas might also be a cause of defective lipid transport.

In addition, we present the thoracoscopic findings of xanthomatous pleuritis. Thoracoscopy

showed diffuse yellow plaque-like deposits in the pleura. This finding was similar to that in a previous report by Bateman et al.³ Xanthoma comprises yellow plaque-like lesions characterized by the presence of lipid-containing histocytes and is commonly found in the gastrointestinal tract.⁵ The endoscopic finding of xanthoma in the gastrointestinal tract includes yellow spots,^{6,7} which is very similar to the finding in our case and a previous report.³ Hence, the yellow plaque-like deposits might be the characteristic finding in xanthomatous pleuritis. Physicians should consider the possibility of xanthomatous pleuritis when such

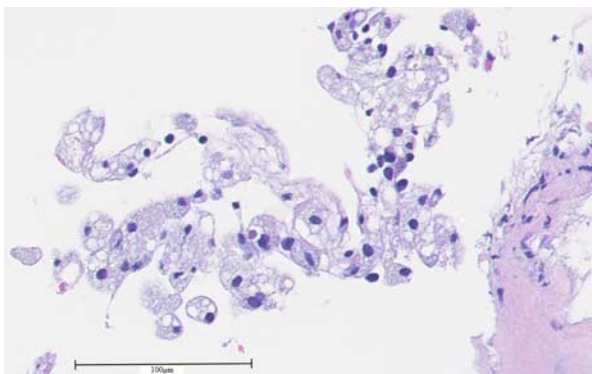


FIGURE 2. Biopsy specimen of the pleura with yellow plaque-like deposit shows foamy cells ($\times 400$ magnification).



FIGURE 3. Endoscopic retrograde cholangiopancreatography reveals meandering pancreatic duct and contrast outflow to the left thoracic cavity (red arrow).

thoracoscopy findings are observed. Furthermore, the thoracoscopy finding of xanthomatous pleuritis may indicate the possibility of the presence of pancreatic disease, even though pancreatic disease is not initially considered as a differential diagnosis when physicians examine patients with pleural effusion. Therefore, abdominal examinations, such as abdominal CT, ultrasound, or endoscopic retrograde cholangiopancreatography, should be performed when physicians confirm this thoracoscopy finding.

Treatments for xanthomatous pleuritis are yet to be established, although steroids have been used in the previous cases.^{1,3} Since xanthomatous pleuritis was considered to be associated with pancreaticopleural fistula in the present case, we performed distal pancreatectomy, which led to a favorable outcome. It may be important to find out the underlying causes, such as pancreaticopleural fistula, when physicians diagnose xanthomatous pleuritis.

In conclusion, the present case suggests that pancreatic pleural effusion might be a cause of

xanthomatous pleuritis, and the thoracoscopic finding of yellow plaque-like deposits in pleura might be the characteristic finding of xanthomatous pleuritis.

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