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Case report

A case of non-specific interstitial pneumonia with recurrent gastric carcinoma and anti-Jo-1 antibody positive myositis

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ABSTRACT

We report the first case of non-specific interstitial pneumonia (NSIP) in a patient with cancer-associated myositis (CAM) that emerged along with the recurrence of the cancer. A 60-year-old woman, with a history of partial gastrectomy for gastric cancer 11 years ago, presented with exertional dyspnea with anti-Jo-1 antibody-positive myositis. Surgical lung biopsy showed NSIP with metastatic gastric cancer. Accordingly, her condition was diagnosed as CAM with cancer recurrence. In patients with a history of cancer, development of myositis may indicate cancer recurrence; therefore, careful observation would be necessary.

1. Introduction

Cancer-associated myositis (CAM) develops before or after a diagnosis of cancer or malignant neoplasm, such as non-Hodgkin's lymphoma, ovarian cancer, pulmonary cancer, breast cancer, or gastrointestinal carcinomas. CAM may sometimes improve after cancer resection [1] or successful chemotherapy. However, cancer progression or recurrence may exacerbate CAM that has initially emerged together with the first presentation of cancer [1–3]. On the other hand, there are only a few cases that report new-onset CAM associated with the recurrence of cancer

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[4–7]. To the best of our knowledge, this is the first report on interstitial pneumonia as a manifestation of CAM that developed for the first time in parallel with cancer recurrence. In addition, this case presented as CAM with Jo-1 antibody, despite the fact that presence of anti-aminoacyl-tRNA synthetase antibodies, like Jo-1, have been known to be associated with myositis without cancer rather than CAM.

2. Case report

A 60-year-old woman presented at our hospital because of a 2-month history of exertional dyspnea with associated morning stiffness of the fingers on both hands. Review of systems revealed gradual muscle weakness on the proximal upper and lower limbs. Eleven years ago, she had undergone partial gastrectomy for signet-ring cell gastric carcinoma, stage T2N1M0, followed by adjuvant chemotherapy. Follow-up surveillance lasting 30 months did not reveal any recurrence. Physical examination at presentation revealed bibasilar end-inspiratory crackles and swelling of the fingers of both hands, with mechanic's hand deformity on the right middle finger and no erythema.

Serum chemistry showed elevated titers of lactate dehydrogenase (LDH; 269 IU/L); creatine kinase (CK; 1,330 IU/mL); aldolase (48.1 U/L); CA19-9 (1590 U/mL); rheumatoid factor (RF; 2.3 IU/mL); antinuclear antibody (ANA; 1:160 homogeneous, 1:160 speckled); and anti-histidyl-tRNA synthetase (Jo-1) antibody (379 U/mL). The value of KL-6 was normal at 317 U/mL. Immunostaining with antibodies against double-stranded DNA, RNP, Sm, SS-A, SS-B, Scl-70, MPO-ANCA, and PR3-ANCA gave negative results.

To investigate the cause of muscle weakness of the upper and lower limbs, magnetic resonance imaging was performed and demonstrated inflammation of the gluteus maximus; electromyography showed a myogenic pattern. Chest radiograph and computed tomography (CT) images showed bilateral infiltrates with predominance in the lower lung fields (Fig. 1). Her pulmonary function test showed restrictive impairment: VC, 1.87 L; VC%, 71.9%; FEV1/FVC, 83.2%; and D_{LCO}/VA, 81.1% of the predicted value. Bronchoalveolar lavage did not reveal specific findings. Transbronchial lung biopsy of the left B^8 and B^{9B} showed granulation tissue in the peripheral air spaces and alveolitis with immature fibrosis.

To obtain pathologic diagnosis of the bilateral lung lesions, lung biopsies from the right S^2 and S^9 segments were performed by video-assisted thoracoscopic surgery. The lung specimens were taken from the right upper and lower lobes (rtS^2 and S^9). On pathologic examination, the biopsy specimens revealed non-specific interstitial pneumonia (NSIP) with an organizing pneumonia (OP) pattern (Fig. 2). Unexpectedly, the intrapulmonary lymph node from the right S^9 lung specimen showed carcinoma cells with histologic features of both signet-ring cell carcinoma and poorly differentiated adenocarcinoma; immunohistochemical staining was negative for thyroid transcription factor-1 (TTF-1), but positive for both cytokeratin-7 (CK7) and cytokeratin-20 (CK20), suggesting metastasis from gastric carcinoma (Figs. 2a and c) [8]. On review, the pathologic features were similar to those of the resected gastric carcinoma 11 years prior (Figs. 2c,e and 3). Furthermore, metastatic foci were found in the lymphatic vessels.

Positron emission tomography (PET) and gastroenterological endoscopy did not reveal any recurrence of the primary tumor, but an abdominal CT showed para-aortic lymph node swelling. Therefore, we diagnosed the patient as having metastatic lymphangitic carcinomatosis of the lung from gastric carcinoma with NSIP and CAM. (Figs. 2 and 3).

The patient was administered oral prednisolone at 50 mg/day, which afforded improvement of the lung interstitial infiltrates and muscle weakness after 1 month. Thereafter, we started gastric cancer chemotherapy with S-1 plus cisplatin. After 1 month, we changed the anticancer drugs to docetaxel because of colon erosion with hemorrhagic stools. After 2 months, docetaxel was changed to S-1 because of elevation of CA19-9 titer to 3204 U/mL. After two more months, however, we stopped chemotherapy because of a persistently elevated and increasing CA19-9 titer at 10910 U/mL. Moreover, a CT scans showed progression of lung infiltrates. It was difficult to determine whether the infiltrates were caused by worsening NSIP or lymphangitis. The myositis did not relapse. However, the patient died 11 months after her initial presentation.

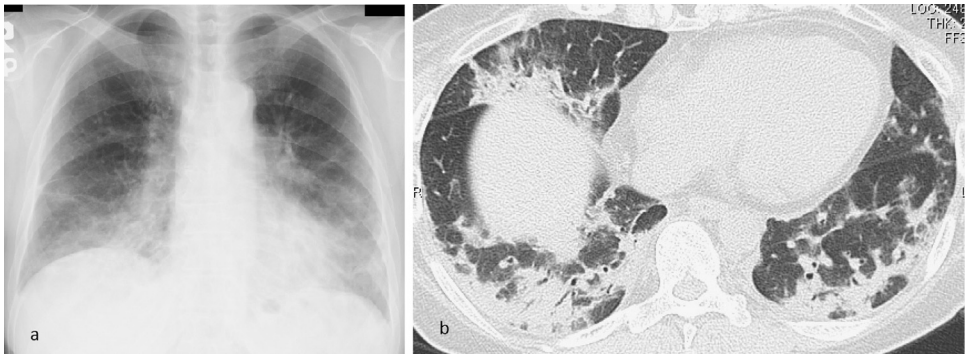

Fig. 1 – Chest radiologic imaging findings in a 60-year-old woman with recurrent gastric cancer. (a) Chest radiograph shows linear and ground-glass shadows in the bilateral lungs, predominantly in the lower lung fields. (b) Computed tomography on admission shows bilateral airspace consolidation, predominantly in the peribronchial area, with irregular margins and areas of ground-glass opacities.