Mikulicz’s disease is a immunoglobulin G4-related systemic disease indicated by enlargement of the lacrimal and salivary glands which differs substantially from Sjögren’s syndrome. A 45-year-old woman who noted bilateral swelling in the submandibular region and dry mouth before 6 month ago was admitted. Ultrasonography of the enlarged glands revealed irregular, hypoechoic and multilobular mass, while the power Doppler showed the presence of hypervascular mass. Laboratory data showed elevated immunoglobulin G level, positive Schirmer’s I test and positive unstimulated whole salivary flow test but was negative for antinuclear antibody, anti-Ro/SS-A antibody and anti-La/SS-B antibody. The biopsy of submandibular gland showed diffuse infiltration of lymphocytes and plasma cells expressing immunoglobulin G4. We report a case of patient diagnosed as Mikulicz’s disease showing immunoglobulin G4 expressing plasma cell infiltration and unique ultrasonographic findings.

A case of primary sjögren's syndrome complicated by bilateral pleural effusion

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Background/Aims: Sjögren's syndrome is a chronic inflammatory autoimmune disorder characterized by lymphocytic infiltration of the exocrine glands resulting in the classical sicca syndrome of dry eyes, dry mouth and salivary gland enlargement. Interstitial pulmonary fibrosis and tracheobronchial sicca are the most common presentations of pulmonary involvement in primary sjögren's syndrome. However, it is rarely accompanied by serositis such as pleuritis or pericarditis. In fact, pleural effusion in primary Sjögren's syndrome has not been reported in Korean patient. We here report a case of bilateral pleural effusion in primary Sjögren’s syndrome with a brief review of the literature. Case: A 61-years-old female patient had complained dyspnea for 3 months and large amount of bilateral pleural effusion was noted on chest radiograph. She underwent pleural fluid analysis, imaging studies including chest and abdominal CT scans, and echocardiogram but there was no evidence of infection, malignancy or cardiovascular disease. She was diagnosed with primary Sjögren’s syndrome by serologic test and sialometry and possibility of other collagen diseases could be excluded from consideration by history and serology. Dyspnea was improved with low dose steroid and NSAIDs and follow-up of imaging also showed reduction of pleural effusion.