Letter to the Editor

Rheumatology

Overlap between Sjogren's syndrome and anti-synthetase syndrome: association or coincidence?

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Sjogren's syndrome is a chronic autoimmune disease characterized by lymphocytic infiltration of the exocrine glands and B-cell dysfunction [1]. In recent years, with the detection of myositis-specific antibodies, a small number of patients with Sjogren's syndrome have been found to be positive for anti-synthetase syndrome (ASS).

A 71-year-old woman presented to the hospital with shortness of breath for 1 month. She was previously in good health, but recently she had frequent dry mouth and a gritty feeling in her eyes, which persisted for 2 years without relief. Physical examination showed significant caries, mechanic's hand and Raynaud's phenomenon, and Velcro rales could be heard in both lower lungs. Laboratory tests demonstrated normal blood cell count and erythrocyte sedimentation rate of 26 mm/h; lactate dehydrogenase, liver function tests, and serum immunofixation electrophoresis were within normal limits. She was positive for anti-nuclear antibody (1 : 320 titer), anti-SSA and anti-Ro-52 antibodies. High-resolution computed tomography showed non-specific interstitial pneumonia (NSIP), and pulmonary function tests suggested a moderate decrease in diffusion function. The salivary flow rate was 0.1 ml/min and biopsy of the labial gland showed focal lymphocytic sialadenitis, which was consistent with the diagnosis of Sjogren's syndrome. Since the patient had progressive shortness of breath, abnormally elevated temperature (maximum body temperature of more than 38.5°C in the absence of infection) and positive anti-Ro-52 antibodies, the possibility of idiopathic inflammatory myopathy could not be ruled out. A myositis antibody test was performed, and anti-OJ antibodies were found to be positive. The patient had no evidence of other connective tissue diseases and was finally diagnosed with primary Sjogren's syndrome combined with ASS according to the appropriate diagnostic criteria [1, 2]. The patient was treated with methylprednisolone 20 mg daily, cyclophosphamide 0.4 g per week and *Tripterygium wilfordii* for 3 weeks, was discharged in good condition with repeat pulmonary diffusion function tests suggesting no deterioration, and is still being followed up.

Sjogren's syndrome can co-occur with a variety of connective tissue diseases. The concept of overlapping myositis (OM) was first introduced in 2006 in the long-term follow-up of patients with idiopathic inflammatory myopathies [3]. In recent years, the medical community has generally recognized ASS as a subtype of overlap myositis [4]. Therefore,

caution should be exercised when giving patients a separate diagnosis of Sjogren's syndrome. The following methods can help further diagnosis. Firstly, Sjogren's syndrome is a systemic autoimmune disease with an interstitial lung disease prevalence of approximately 20% [5]. It has been shown that the severity of Sjogren's syndrome-associated interstitial lung disease (ILD) is usually lower than that of ILD associated with other rheumatic diseases. The diagnosis of Sjogren's syndrome alone should be considered if the interstitial lung disease is severe or rapidly progressive [6]. Secondly, the presentation of ILD on high-resolution computed tomography can help to identify Sjogren's syndrome or combined ASS. It is generally recognized that the most common type of ILD in Sjogren's syndrome is NSIP, whereas in idiopathic inflammatory myopathy, cryptogenic organizing pneumonia (COP), NSIP and COP + NSIP predominate [7]. Thirdly, although anti-SSA antibodies (including anti-Ro-52 and anti-Ro-60 antibodies) are specific for the diagnosis of Sjogren's syndrome, anti-Ro-52 antibodies are also myositis-associated antibodies [8]. Studies have shown that ASS patients with positive anti-Ro-52 antibodies have a higher incidence of ILD and mortality than those without anti-Ro-52 antibodies, suggesting that anti-Ro-52 antibodies may be the causative antibodies [8]. Thus, the presence of anti-SSA antibodies may indicate idiopathic inflammatory myopathy. Finally, unexplained hyperthermia is less common in Sjogren's syndrome patients during the disease, while it is more common in ASS [9].

In conclusion, among rheumatic diseases, interstitial lung disease is most frequently seen in systemic sclerosis, followed by idiopathic inflammatory myopathies [10]. Early detection of myositis antibodies is recommended for patients with Sjogren's syndrome who present with rapidly progressive interstitial lung disease or hyperthermia.

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Conflict of interest

The authors declare no conflict of interest.

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