be drawn with Hashimoto’s thyroiditis in which almost all patients have thyroid peroxidase Abs, but the initiation of thyroid destruction is believed to be T-cell mediated. In conclusion, clinicians should be aware of a possible auto-immune cause for hypoparathyroidism in patients with SS.

**Rheumatology key message**

- CaSR Abs may define hypoparathyroidism in SS.

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**Anti-synthetase syndrome positive for anti-isoleucyl-tRNA synthetase antibodies: an unusual case overlapping with systemic sclerosis and Sjögren’s syndrome**

Sir, Anti-synthetase syndrome (ASS) is characterized by inflammatory myositis associated with interstitial lung disease and anti-synthetase antibodies [1]. Other symptoms, including arthritis, RP and mechanic’s hands also belong to ASS [2]. Several anti-synthetase antibodies have been described; anti-Jo1 being the most common. Very little is known about the clinical manifestations of the ASS associated with anti-isoleucyl-tRNA synthetase antibodies (anti-OJ antibodies), most likely because anti-OJ antibodies are particularly rare (<3% of myositis) and have not been routinely investigated in the past. Only 18 patients have been reported in the English literature [3–6]. We report an unusual patient with anti-OJ-positive ASS overlapping with SSC and SS.

A 21-year-old African woman was admitted with digital ulcers. She complained of recent RP, gastro-oesophageal reflux, brief arthralgia and dry eyes. Clinical examination and chest X-ray were normal. However, over the next 2 months, she developed a progressive dyspnoea associated with muscle weakness (graded as 3+/5 by manual muscle test). Pulmonary examination revealed fine crackles. A high-resolution chest (CT) scan showed diffuse interstitial pneumonia, with a predominant honeycomb pattern (usual interstitial pneumonia) and pulmonary function tests revealed a restrictive syndrome [vital capacity (VC) = 26%] and carbon monoxide transfer factor (TL_{CO}) was dramatically decreased (26%). Muscular investigations confirmed severe myositis: creatine phosphokinase = 11 700 IU/l (normal range <160 IU/l) and electromyography showed a myogenic syndrome. Muscle biopsy showed endomysial infiltrates with a majority of B cells without vasculitis; immunohistochemistry also showed diffuse MHC class I over-expression, but was negative for complement terminal membrane attack...
complex C5b9. Echocardiography was normal. Finger capillaroscopy disclosed a few megacapillaries. Salivary gland biopsy revealed a focal lymphocytic sialadenitis (Chisholm and Mason score Grade III). CRP was normal, haemoglobin was 9.9 g/dl (minor β-thalassaemia). Electrophoresis demonstrated polyclonal hypergamma-globulinaemia (22 g/l). IF analysis on HEP-2 cells (Bio-Rad, Marnes-La-Coquette, France) was positive for ANAs (speckled and nucleolar 1/2560). No cytoplasmic pattern was described. Anti-extractable nuclear antibodies were positive for anti-Ro/SSA-60 kDa and anti-La/SSB. Immuno-DOT analysis (Dtek DiaSorin, France) was positive for anti-OJ antibodies. All other autoantibodies, including RF, ANCAs, were negative.

Immunosuppressive treatment was started with a combination of steroids (1 mg/kg/day) and monthly pulses of CYC 0.7 g/m². Steroids were progressively withdrawn and CYC (total of six pulses) was replaced by AZA (2 mg/kg/day). After 41 months of follow-up, the patient demonstrated muscular recovery (normal strength and creatine kinase levels) and pulmonary improvement (stable chest CT scan and increased VC = 46% and TLCO = 48%). However, gastro-oesophageal reflux worsened concomitantly with the development of a sclerodermic oesophagus on repeated chest CT scan. The patient also developed sclerodactyly (Rodnan score = 20) and poikiloderma without telangiectasia or mechanic’s hands. Severe RP persisted despite the use of calcium inhibitors and the patient’ digital ulcers relapsed every winter, requiring specific vascular treatments.

ASS is a heterogeneous disease involving numerous organs. The patient reported herein illustrates the broad spectrum of the disease, with the involvement of five organs, including lung, muscle, oesophagus, microvasculature and skin. The relative frequency of different organ involvement varies among patients with ASS, and could be related to anti-synthetase antibody subtypes. Thus, some have reported lower frequencies of both myositis and RP in association with anti-OJ antibodies [4, 6] compared with anti-Jo1 antibodies. However, the current patient presented with both myositis and RP as the most severe disease manifestations.

Clinical symptoms of ASS may overlap with other CTDs, including SS and SSc [1, 2, 7, 8]. However, such patients rarely fulfill the classification criteria for these diseases [9, 10]. Conversely, the current patient seemed original as she did fulfill the ACR criteria for both SSc (RP, megacapillaries and Rodnan score ≥ 14) and primary SS (dry eyes, positive Shirmer test, focal lymphocytic sialadenitis and anti-Ro/SSA-60 kDa and anti-La/SSB). Moreover, she disclosed predominant SSc symptoms, which surprisingly evolved and worsened under treatment, in contrast to her muscular and pulmonary involvement.

The current patient was diagnosed with anti-OJ-positive ASS, and was remarkable for presenting with predominant signs of SSC and SS. This is in contrast to previous studies, and underlines the individual heterogeneity of ASS that exists within antibody subtype.