Abstract

Ankylosing spondylitis is reported to involve not only 
the joints but neurologic systems as well. The association 
of MS and AS has rarely been reported in the literature 
and epidemiological studies did not prove a definite 
relationship between these two conditions at present.

We here describe a HLA-B 27 positive AS patient with 
MS symptoms and review the literature on the association 
of two diseases.

Key words: Ankylosing spondylitis; multiple sclerosis; 
neurological complications.

Introduction

Ankylosing spondylitis (AS) is a chronic inflammatory rheumatic condition, primarily affecting 
the axial skeleton and large peripheral joints. The disease is characterized by low back pain, limitation 
of spinal mobility and radiological evidence of sacroiliitis and spondylitis, and can be associated 
with extra-articular involvement including several neurological symptoms (1). The neurologic complications of AS are rare and include single root lesions, monophasic myelopathy, spinal cord compression 
due to atlanto-axial subluxation or fracture of stiff vertebrae, cauda equina syndrome and rarely multiple sclerosis (MS) or MS-like syndrome (2).

The cause of MS remains unknown. There is evidence for an environmental trigger coupled with 
genetic susceptibility contributing to MS development (3). The association of MS and AS has rarely 
been reported in the literature and epidemiological studies did not prove a definite relationship between 
these two conditions at present (4).

We report a HLA-B 27 positive AS patient with 
MS symptoms and review the literature on the association of these two diseases.

Case Report

A 34-year-old man known with a 15-year history 
of AS; presented to the outpatient clinic of physical medicine and rehabilitation with difficulty in walking and balance, increased leg muscle tone and fatigue. He also complained of difficulty voiding, constipation and decreased libido. He mentioned that his symptoms had begun more than one year ago but had progressed over the last 6 months. His medical history was positive for uveitis, left total hip arthroplasty 5 years ago and upper gastrointestinal tract bleeding 4 months ago. He was on sulfasalazin 2 g/day and 150 mg/day indomethacine with a proton pump inhibitor. He denied having fever. He was not smoking and not drinking alcohol.

On physical examination his vital signs were normal. Mobility of lumbar and thoracic spine, bilateral hips and shoulders was markedly reduced and he had a kyphotic posture. Radiologic examination revealed grade 4 sacroiliitis and complete ankylosis of the dorso-lumbar spine. Modified Schober test was 1 cm, a chest expansion test was 2 cm. The distance from 
ground to finger was 40 cm, the wall to tragus 36 cm, the distance between two malleoli was 47 cm. 

General joint pain intensity by VAS was 40 cm. The Bath Ankylosing Spondylitis Disease Activity Index 
(BASDAI), The Bath Ankylosing Spondylitis Metrology Index (BASMI) and the Bath Ankylosing 
Spondylitis Functional Index (BASFI) were 3.8, 8 and 96 respectively. The neurological examination 
revealed hyperactivity of all reflexes and positivity of pathologic reflexes. Bilaterally spastic paraparesis in lower extremities and an intentional tremor of upper extremities were recorded. There was no sensational abnormality and Romberg test was negative.

Routine complete blood, biochemical tests, and 
tests for tumour markers, vitamin B12 level and
thyroid functions were in normal ranges. Erythrocyte sedimentation rate and C-reactive protein were 64 mm/h and 8.35 mg/dl respectively. Serological tests for Brucella, HIV and hepatitis were negative. HLA-B27 was positive.

There was microscopic hematuria in complete urine analysis. Urodynamic evaluation showed hyperkinetic neurogenic bladder.

Lumbar puncture (LP) could not be performed due to the limitation and ankylosis of the lumbar spine. Pelvic X-ray showed bilateral sacroileitis and total hip arthroplasty in the effect side (Fig. 1). Brain Magnetic Resonance Imaging (MRI) showed multiple foci of increased signal intensity in the periventricular and subcortical white matter bilaterally (Fig. 2). Spine MRI showed focal centromedullar high intensity at C4-5 (Fig. 3), suggesting a demyelinating disease process. Visual evoked potential (VEP) latencies were prolonged bilaterally (P100 latency for the right eye: 128.1 msec, for the left eye: 127.8 msec), somatosensory evoked potential (SEP) latencies were prolonged at the right tibial posterior nerve (P1: 45.8 msec, P2: 66.9 msec) and absent at left and right tibial posterior nerves (P1:0, P2:0). The third wave of the brain stem auditory evoked potential (BAEP) was also prolonged (right III: 4.00 msec, left III: 4.24 msec) on both sides.

**Discussion**

A number of AS cases presenting with possible or definite MS have been reported in the English literature (5-12). Libbrecht and De Bleecker (7) reviewed the literature for the association of AS and MS or MS-like conditions and reported 9 cases before their case, in which only 4 of them had definite MS. Since then, 6 cases were reported and only 3 had a diagnosis of definite MS (Table 1). This illustrates that the association of AS and definite MS is very rarely reported in the literature. In all reported cases, AS preceded the first signs of MS. The intervals between two conditions ranged from 3-21 years. In most of the earlier cases, information was missing about clinical, radiologic or cerebrospinal fluid (CSF) findings consistent with MS, due to the
Degeneracy. Activation of T lymphocytes is concerned in both diseases, although there is no evidence of a common favoring factor and the antigenic stimulus remains unknown for both diseases (7). Absence of complete concordance of disease expression of both AS and MS suggests that environmental factors may play a role in both diseases but still the etiopathogenesis of the association of these two diseases needs to be illuminated (14).

Sufficient evidence does not seem to be available to support a true association of AS and MS. Pillay and Hunter reported an increased prevalence of abnormal evoked potentials in AS patients suggesting an association with MS-like disease and AS (15). On the other hand, Hanrahan et al. (6) investigated the prevalence of HLA-B27 in patients with MS and indicated not only a higher incidence of HLA-B27 positivity but also sacroiliitis. Although they could not indicate an abnormality in VEP, BAEP and SEP except some minor peripheral abnormalities in their study group, these results may also indicate a strong association between AS and MS. We believe that future studies will bring vigorous evidence highlighting the association of these two conditions.

In conclusion, definite MS may be a separate neurological manifestation associated with AS and the coexistence of AS and MS can occur at a much higher incidence than might be expected. Future studies are needed for sufficient evidence indicating the relationship between these suffering diseases.
REFERENCES


Figen Tuncay, M.D.,
Associate Professor,
Ankara Training and Research Hospital,
Clinic of Physical Medicine and Rehabilitation,
Ankara, Turkey.
E-mail: figengokoglu@mynet.com