

Case Reports

Ankylosing Spondylitis with Peripheral Neuropathy - A Rare Case Report

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Abstract:

Ankylosing Spondylosis (AS) is a chronic inflammatory disorder of joints predominantly affecting the sacroiliac joints and spine. Besides the joints, it affects many extra articular sites like heart, eye, lungs etc. Neurological complication is very rare in AS. Here we report a case of a 30 years old man of AS with peripheral neuropathy.

Key Words: Ankylosing Spondylitis, Peripheral Neuropathy.

Introduction:

Ankylosing Spondylitis (AS) is an inflammatory disorder of unknown etiology that primarily affects the axial skeleton; peripheral joints and extra articular structures are also frequently involved. The disease usually begins in the second or third decade; male to female ratio is 3:1. Common sites of joint involvement are sacroiliac joints, spine, ischial tuberosities and heels.¹ The most common extra articular manifestation is anterior uveitis (40%). Aortic insufficiency and Inflammatory Bowel Disease may also be associated with AS. Neurological complication is very rare & occurs late in the course of the disease.

Case Report

A 30-year-old married, Muslim, non-smoker, non-alcoholic, non-diabetic and normotensive gentleman has been suffering from pain & swelling in both the ankle & knee joint for ten years. The pain increases during rest and in the morning

and subsides to some extent after activity as the day progresses. He took different NSAIDs for a long time which suppressed the pain for a while. He received corticosteroid for variable period for several occasions. In addition he experienced pain in the low back region and neck for 5 years. Initially it was mild in nature with morning stiffness for few minutes, not hampered his daily activities. But for the last 5 months it has increased in severity with morning stiffness for 30 minutes, relieves with activity without radiation. It was associated with pain in the left heel pad while walking. One month later he developed pain and swelling of left ankle which was insidious in onset with restricted movement. He also noticed tingling & numbness of hands and feet for last 5 months which is increasing gradually over last 2 months. This was not associated with speech difficulty, cognitive impairment, bowel bladder involvement. It has no definite aggravating or relieving factor. He also noticed pain in the left thumb base, wrist, elbow & shoulder joint for the last 2 months with morning stiffness which relieves by activity. He denies any history of conjunctivitis, urethritis, diarrhoea or rash. He denies any history of unprotected extramarital sex. Though he works in a leather industry he gives no history of contact with toxic chemicals. He is not immunized; comes from a low middle class family.

On examination he was found anxious & ill-looking, mildly anemic, pulse - 84 beats/min, B.P - 110/70 mm of Hg. Left ankle was swollen, tender (grade 2), local temperature raised, movement restricted in all directions. The spine was found normal. Schober's test was negative.

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Tenderness over sacro iliac joint was present. Higher psychic function including speech was normal. Cranial nerves including fundus were normal. There was wasting of calf muscles of both lower limb and also of thenar & hypothenar muscles of left hand & thenar muscles of right hand. Muscle tone was normal. Muscle power of upper limb - 4/5 (right) & 3/5 (left), lower limb - 4/5 (right) & 3/5 (left). Right and Left Biceps, Triceps, Supinator and Knee jerks are diminished. Both ankle jerks were absent, plantar response was equivocal on both side. There was impaired sensation in gloves & stocking pattern. Coordination was impaired due to weakness. There was no cerebellar sign and gait was normal. Investigation report revealed ESR - 121 mm in 1st hour, Hb - 9.3 gm/dl, total count of WBC - 13,100 /cmm. On differential count neutrophil was 69.7 %, Lymphocyte 9.6 %, M - 6.4 %, E - 4.3 %, RBC - 4.35 million/cmm, Platelet count - 8,39,000 /cmm, MCV - 72 fl, MCH - 21 pg, MCHC - 30 gm/dl. Peripheral blood film revealed microcytic hypochromic anemia with anisopoikilocytosis, WBC series was normal in count & matured into above distribution, Platelet count was normal. Comment - suggestive of hereditary haemoglobin disorder. Urine R/M/E - Normal. Serum ferritin - 265 microgm /L. Hb electrophoresis showed Hb A - 72.45 %, Hb E - 27.55 %, Comment - Hb - E trait. RBS - 6.2 mmol /L. Serum uric acid - 4.7 mg /dl. Serum creatinine - 0.86 mg /dl. Xray chest P/A view - normal. X - Ray lumbosacral spine B/V - normal. X - ray Sacro-iliac joint shows irregularity & loss of cortical margin and sclerosis .



Fig-1: *Bilateral sacroiliitis*

X-ray cervical spine B/V was normal. X-ray left ankle was normal. RA test, Anti CCP antibody and ANA were negative, CRP was high (109 mg /L), HLA-B27 was positive, ECG tracing revealed incomplete RBBB; Echocardiogram was normal. NCS of left lower limb & right upper limb suggests chronic demyelinating polyneuropathy with secondary axonal involvement. EMG reveals features of denervation with some features of reinnervation in some of the sample muscle.

Discussion

Patients with a chronic inflammatory arthritis including AS with peripheral neuropathy is a rare condition. Briefly, Ankylosing spondylitis is a chronic inflammatory disease of the joints of the axial skeleton² predominantly affecting the sacroiliac joints and spine¹ manifested clinically by pain and progressive stiffening of the spine.³ The onset is typically between the ages of 20 and 30, with a male preponderance of about 3:1. HLA B27 is positive in 90 % of patients with AS. The overall prevalence is less than 0.5 % in most populations.¹ It is thought to arise from an as yet ill defined interaction between environmental pathogens and the host immune system in genetically susceptible individuals.¹ On the other hand neuropathic pain is a positive neurological symptom caused by dysfunction of the pain perception apparatus, in contrast to nociceptive pain, which is secondary to pathological processes such as inflammation.² The most common causes of neuropathic pain are diabetic neuropathies, trigeminal and post herpetic neuralgias, trauma to a peripheral nerve.² Single nerve lesions cause disturbance in the sensory distribution of the nerve, whereas in diffuse neuropathies the longest neurons are affected first, giving a characteristic 'glove and stocking' distribution². Possible causes of neuropathy may be mechanical compression of nerves by swelling of soft tissue, bone erosions and joint deformity.⁴ However, it is often difficult to diagnose slight or early neuropathies with any certainty, and the study of the peripheral neuromuscular system is made difficult by symptoms resulting from pain and stiffness of peripheral joints.^{5,6} Patients with evident joint pain can describe additional symptoms like muscle weakness and paresthesia that can suggest neuropathy.⁷⁻¹⁰ The peripheral nervous system can as well be involved as the central nervous system in asymptomatic AS patients.⁵ On June, 12, 2014 : 18,758 people who have AS are studied. Among them, 8 (0.04 %) have Neuropathy (Peripheral). They amount to 0.03 % of all the 26,447 people who have Neuropathy. Trend of Neuropathy in AS reports in 2006 - 2 people, in 2007 - 5 people & in 2014 - 1 person. Gender of people who have AS and experienced Neuropathy: Female - 33.33 %, Male - 66.67 %. Age of people who have AS and experienced Neuropathy : 0 - 1 year - 0.00 %, 2 - 9 years - 0.00 %, 10 - 19 years - 0.00 %, 20 - 29 years - 0.00 %, 30 - 39 years - 66.67 %, 40 - 49 years - 0.00 %, 50 - 59 years - 33.33 %, 60 + years - 33.33 %. Severity of the symptom : (Neuropathy) - least (0.00 %), moderate (100.00 %), severe (0.00 %), most severe (0.00 %).³ An accurate diagnosis is therefore possible when the characteristic

changes of both diseases are present along with the investigation. Our case satisfies diagnostic criteria of both the diseases. Although very rare yet we may encounter such a case seldom in our clinical practice. As a result, we conclude that peripheral nerve involvement is one of the rare striking extra-articular involvements of AS, with no apparent correlation with the clinical parameters.

Conflict of Interest : None

References

1. Fauci Anthony S, Kasper L, Dennis et al. Harrison's Principles of Internal Medicine. 2008;17(2):2109-12.
2. Brian R. Walker, Nicki R. Colledge, Stuart H. Ralstone, Ian D. Penman; Davidson's Principle and Practice of Medicine. 2014;26:1105,1164-65.
3. Hellmann DB, Imboden JB. Current Medical Diagnosis & Treatment 2014;20:822.
4. eHealthMe FDA and social Media, personalized in Jun, 12, 2014 .
5. Nerve conduction studies in patients with ankylosing spondylitis. J Natl Med Assoc. 2010; 102(3):243-6 .
6. Bekklund S, Torbergsen T, Omdal R, Husby G, Mellgren SI. Nerve conduction studies in rheumatoid arthritis. Scand J Rheumatol. 1996;25:287-92.
7. Sivri A, Uysal GF. The electroneurophysiological findings in rheumatoid arthritis patients. Electromyogr Clin Neurophysiol. 1999;39:387-91.
8. Lang AH, Kalliomaki JL, Puusa A, Halonen JP. Sensory neuropathy in rheumatoid arthritis: an electroneurographic study. Scand J Rheumatol. 1981;10:81-4.
9. Bekkelund SI, Mellgren SI, Proven A, Husby G. Quantified neurological examination with emphasis on motor and sensory functions in patients with rheumatoid arthritis and controls. Br J Rheumatol. 1996;35:1116-21.
10. Lanzillo B, Pappone N, Crisci C, di Girolamo C, Massini R, Caruso G. Subclinical peripheral nerve involvement in patients with rheumatoid arthritis. Arthritis Rheum. 1998;41:1196 – 202.