Axonal Sensory-Motor Polyneuropathy in Ankylosing Spondylitis: A Case Report

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Introduction

Ankylosing spondylitis (AS) is an inflammatory disorder with many unidentified reasons. The primary involvement in AS cases is an axial skeleton, defined by joint deformity, stiffness, enthesitis with pain, and sacroiliac arthritis (1). Systemic inflammation leads to stiffness and pain in the entheses, spine, and sacroiliac joint resulting in spinal curvature loss and movement restriction. Advanced disease is possible to lead to spine and sacroiliac joint fusion, causing a "bamboo spine" condition. Enthesopathy, including plantar fasciitis and Achilles tendonitis, may happen early stage of the disease and result in structural damage. Peripheral joints, primarily in the lower limbs such as the hip, and knee, can also be affected (2).

Neuropathic pain can be experienced in AS cases, endorsed by abnormalities in the brain's gray matter and neural correlates. The AS clinical picture includes neuropathic pain, mood deficits, and motor impairment. Hence, back pain in subjects with AS may be related to neuropathic pain (3). Also, in AS cases, no precise data showed Axonal Sensory-Motor Polyneuropathy in these patients. Although, some research found a correlation between AS and peripheral neuropathy (3-5). Thus, this case report revealed an AS subject with polyneuropathy, a rare disease manifestation.

Case Presentation

A 45-year-old male with no prior medical history was diagnosed with AS about ten years ago following bilateral Achilles tendonitis, rupture of the left Achilles tendon, inflammatory low back pain, positive HLA B27, and increased inflammatory markers. Sacroiliac X-ray showed bilateral Sacroiliitis (Figure 1), then he was managed with Sulfasalazine and NSAID at anti-inflammatory. About one year ago, the case felt tingling and numbness, firstly in the upper limbs and then in the lower limbs. Upon consultation with a rheumatologist, he underwent tests such as EMG-NCV, vitamin B12, and folate levels. EMG-NCV result showed axonal-type sensory-motor polyneuropathy and laboratory tests demonstrated a normal range of vitamin B12 and folate levels. Laboratory tests showed in Table 1.

Considering the inconsistency between the patient's neuropathy and AS, additional examinations were performed, including CT scans of the lungs, abdomen, and pelvis and tumor marker tests to investigate the possibility of malignancy. The tumor markers range is shown in Table 1. All requested tests demonstrated normal results, and the criteria for demyelinating neuropathy were absent in the EMG-NCV; this neuropathy subtype was not considered a potential etiology. Eventually, based on the findings, no justification for the patient's neuropathy was identified, and the absence of any secondary cause was proposed as the etiology; hence, AS considers an etiology of Axonal Sensory-Motor Polyneuropathy, a rare presentation for this disease.

Discussion

0.1–0.5% of people are affected by AS, and it is defined by radiographic sacroiliitis, inflammatory back pain, excess spinal bone formation, and positive HLA-B27 in most cases (6, 7). AS spondyloarthropathy features are associated with human leukocyte antigen (HLA)-B27 (8, 9). Positive HLA-B27 AS patients are susceptible to younger AS cases, have a higher uveitis frequency, better response to management with tumor necrosis factor-alpha (TNF-α) inhibitors, and have a lower IBD or psoriasis prevalence (10).

Pain is the AS main symptom known as one inflammatory pain (IP) (11). Although, AS subjects' pain always does not relate to the inflammatory indexes, including erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) (12).

Chronic inflammatory arthritis patients like AS with peripheral neuropathy is an infrequent condition. Peripheral nerve involvement is one extra-articular rare striking AS involvement, and maybe it has no association with the clinical variables (13). Furthermore, in asymptomatic AS patients, the involvement of the peripheral nervous system can be the central nervous system (4).

A case-report study showed a case with swelling in his knees and ankles, chronic pain, and neck and low back pain. The patient additionally had tingling and numbness in his hands and feet. On examination, he has impaired sensation, muscle wasting, weakness, absent ankle jerks, and equivocal plantar response. EMG suggests chronic demyelinating polyneuropathy with secondary axonal involvement. Additionally, the patient has a positive HLA-B27 and microcytic hypochromic anemia (13). Their study involved patient who, like ours, had developed neuropathy, but the type of neuropathy differed.

In addition, another study investigated the relationship between peripheral neuropathy and AS by evaluating the peripheral nervous system of 32 AS patients without symptoms of neuropathy. The study found that 18.8% of the AS patients had sensory or sensorimotor peripheral nerve involvement, and 21.9% had focal nerve involvement. The study also found correlations between tibial nerve motor conduction velocity and Schober and chest expansion tests and negative correlations between sural nerve sensory action potential amplitude and age and disease duration. The study suggests that asymptomatic AS patients can have peripheral nervous system involvement, and further studies are needed to confirm the results and evaluate the clinical significance (4). The results of their study were in line with ours, but their cases did not have any symptoms of neuropathy, while ours did.

In summary, our case report presented a case of AS under medication with a rare peripheral neuropathy presentation. Our case report differs from previous studies and case reports.

References

1. Zong HX, Xu SQ, Tong H, Wang XR, Pan MJ, Teng YZ. Effect of anti-tumor necrosis factor α treatment on radiographic progression in patient with ankylosing spondylitis: A systematic review and meta-analysis. Modern rheumatology. 2019;29(3):503-9.

2. Ebrahimiadib N, Berijani S, Ghahari M, Pahlaviani FG. Ankylosing Spondylitis. Journal of ophthalmic & vision research. 2021;16(3):462-9.

3. Wu Q, Inman RD, Davis KD. Neuropathic pain in ankylosing spondylitis: a psychophysics and brain imaging study. Arthritis and rheumatism. 2013;65(6):1494-503.

4. Gündüz OH, Kiralp MZ, Ozçakar L, Cakar E, Yildirim P, Akyuz G. Nerve conduction studies in patients with ankylosing spondylitis. Journal of the National Medical Association. 2010;102(3):243-6.

5. Erdal A, Gündüz OH, Duruöz T. AB0759 Peripheral Nervous System Involvement and Neuropathic Pain in Ankylosing Spondylitis. Annals of the Rheumatic Diseases. 2015;74(Suppl 2):1152-3.

6. Wang R, Ward MM. Epidemiology of axial spondyloarthritis: an update. Current opinion in rheumatology. 2018;30(2):137-43.

7. van der Linden S, Valkenburg HA, Cats A. Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York criteria. Arthritis and rheumatism. 1984;27(4):361-8.

8. Howe HS, Zhao L, Song YW, Springer L, Edmonds J, Gu J, et al. Seronegative spondyloarthropathy–studies from the Asia Pacific region. Annals of the Academy of Medicine, Singapore. 2007;36(2):135-41.

9. Akassou A, Bakri Y. Does HLA-B27 Status Influence Ankylosing Spondylitis Phenotype? Clinical medicine insights Arthritis and musculoskeletal disorders. 2018;11:1179544117751627.

10. Akkoc N, Yarkan H, Kenar G, Khan MA. Ankylosing Spondylitis: HLA-B*27-Positive Versus HLA-B*27-Negative Disease. Current rheumatology reports. 2017;19(5):26.

11. Lindstrom U, Bremander A, Haglund E, Bergman S, Petersson IF, Jacobsson LT. Back pain and health status in patients with clinically diagnosed ankylosing spondylitis, psoriatic arthritis and other spondyloarthritis: a cross-sectional population-based study. BMC musculoskeletal disorders. 2016;17:106.

12. Bidad K, Gracey E, Hemington KS, Mapplebeck JCS, Davis KD, Inman RD. Pain in ankylosing spondylitis: a neuro-immune collaboration. Nature reviews Rheumatology. 2017;13(7):410-20.

13. Biswas S, HAM NA, Biswas PK, Rashid MHU, Sarkar PK, Chowdhury MK, et al. Ankylosing Spondylitis with Peripheral Neuropathy-A Rare Case Report. Journal of Medicine. 2016;17(1):36.

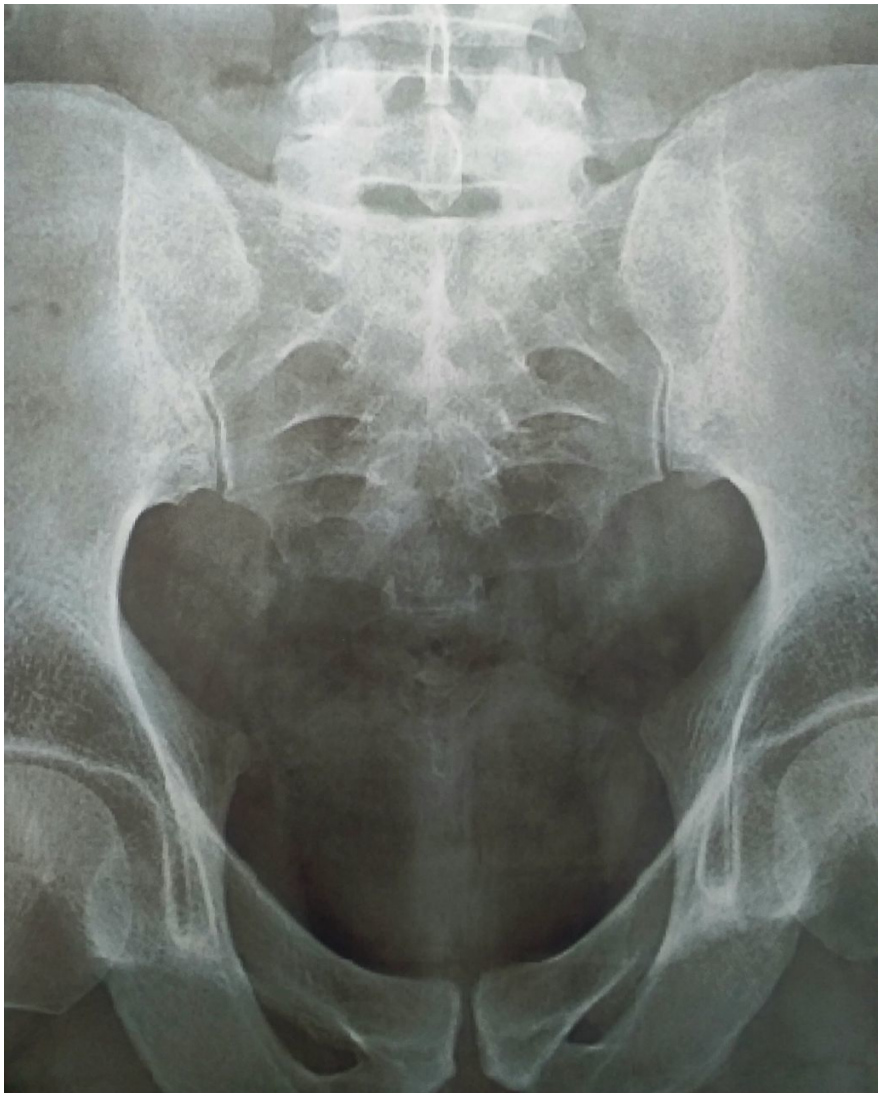

Figure 1. X-ray of the Sacroiliac joint shows bilateral sacroiliitis

Table 1. Laboratory parameters of the patient

Laboratory parameters	Patient's values	Normal range
Leukocyte count, per μL	6.1×10^3	4-10×103
Hemoglobin, g/dL	14.4	12.3-15.3
MCV	93	80-100
Platelet count, per μL	300000	150000-450000
ESR, mm/h	7	0-30
CRP, mg/L	1	< 6
Creatinine, mg/dL	1	0.7-1.3
Na, mmol/L	138	136-145
K, mmol/L	4.5	3.5-5.2
Vitamin D, ng/mL	40	50-70
Vitamin B12, pg/mL	290	187-883
Folic acid, ng/ml	15	3-17
Ferritin, ng/ml	28	21-274
TSH, mIU/L	3.7	0.5-5.0
CA-125, U/mL	6	<21
CA19-9, IU/mL	8	<40
CEA, ng/mL	102	<602
AFp, IU/mL	2	<505

Na: Sodium, K: Potassium, TSH: Thyroid stimulating hormone, CA: Cancer antigen, CEA: Carcinoembryonic antigen, AFp: Alpha-fetoprotein