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CASE REPORT

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Axonal sensory-motor polyneuropathy in ankylosing spondylitis: A case report

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Key Clinical Message

In ankylosing spondylitis cases, axonal-type sensory-motor polyneuropathy is a rare manifestation and should be considered an underlying etiology in patients with unexplained neuropathy.

Abstract

This case report discusses a 45-year-old male diagnosed with ankylosing spondylitis (AS), a chronic inflammatory disorder affecting the axial skeleton and peripheral joints. The patient presented with polyneuropathy, characterized by tingling and numbness in the upper and lower limbs, which is an uncommon manifestation of AS. After undergoing various tests, including CT scans and EMG-NCV, no secondary cause for the neuropathy was identified; AS was considered the etiology of the patient's axonal-type sensory-motor polyneuropathy.

KEYWORDS

ankylosing spondylitis, axonal sensory-motor, polyneuropathy

1 | 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 in the 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 manifestation of AS.

2 | CASE PRESENTATION

A 45-year-old male with no prior medical history was diagnosed with AS about 10 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, and then he was managed with sulfasalazine and NSAID. About 1 year ago, the case felt tingling and numbness, first 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 are shown 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 explanation for the patient's neuropathy was identified, and the absence of any secondary cause was proposed as the etiology; hence, AS is considered an etiology of axonal sensory-motor polyneuropathy, a rare presentation for AS.

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	300,000	150,000–450,000
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

Abbreviations: AFp, alpha-fetoprotein; CA, cancer antigen; CEA, carcinoembryonic antigen; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate; K, potassium; Na, sodium; TSH, thyroid-stimulating hormone.

3 | DISCUSSION

Approximately, 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 HLA-B27.[8,9] AS patients with positive HLA-B27 are susceptible to younger AS cases, have a higher uveitis frequency, better response to management with tumor necrosis factor-alpha inhibitors, and lower IBD or psoriasis prevalence.[10]

Pain is the AS main symptom known as inflammatory pain.[11] However, AS subjects' pain always does not relate to the inflammatory indexes, including erythrocyte sedimentation rate and C-reactive protein.[12]

Peripheral neuropathies can be categorized into polyneuropathies, multifocal neuropathies, and mononeuropathies, which are peripheral nervous system diseases. Generally, paresthesia and numbness are peripheral neuropathy symptoms. Usually, these symptoms are with pain and weakness. Polyneuropathies are classified into demyelinating forms and axonal types, which are vital for diagnostic causes. More than half of cases with peripheral neuropathies progress over months or years, while some have rapidly developed.[13]

Peripheral neuropathy's pathophysiology with usual mechanisms is determined by primary causes or diseases, including axonal degeneration, Wallerian degeneration, and segmental demyelination. Wallerian degeneration arises from nerve axon damage, leading to the distal portion wasting away because of nutrient deprivation. This results in focal mononeuropathy, generally caused by infarction or trauma. Myelin sheath degeneration is affected by segmental demyelination, however, sparing the nerve axon. It happens frequently in immune-mediated and inflammatory conditions, causing motor and sensorimotor neuropathies.[14,15]

The dying-back phenomenon, or axonal degeneration, commonly shows symmetrical polyneuropathy with weakness, mainly in foot dorsiflexion and ankles, followed by muscle trophic alternations. This degeneration initiates from the distal and then advances to the proximal due to the axon's distant part being far from the cell body. It does not receive as much energy and other resources from the cell body as the parts of the axon that are closer to the cell body. Suggested mechanisms include enhanced intra-axonal calcium levels and impaired axonal survival factors delivery, which causes calcium-dependent cytoskeletal breakdown.[16,17]

In adults, polyneuropathies, or peripheral neuropathies, are the most frequent peripheral nervous system disorders, particularly in the elderly, with a 5%–8% estimated prevalence.[18] Diabetes is the most frequent

polyneuropathy etiologies in North America and Europe. Among chronic alcoholism people, alcohol-associated polyneuropathy has a 22%–66% prevalence. Polyneuropathy may result from various immunological processes, vitamin deficiency or overdose, exposure to toxic substances and drugs, and genetic reasons. Approximately 50% of cases with polyneuropathy are correlated with pain.[18] One of the AS remarkable extra-articular is peripheral nerve involvement, with no associated clinical variables.[19]

A case report showed a case with swelling in his knees and ankles, chronic pain, and neck and lower back pain. The patient additionally had tingling and numbness in his hands and feet. On examination, the patient 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.[19] Their study involved a patient who, like ours, had developed neuropathy, but the type 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.[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 conclusion, AS primarily affects the spine and sacroiliac joints, causing pain and stiffness. This case report shows a rare manifestation of AS—axonal sensorymotor polyneuropathy. A 45-year-old patient with AS developed limb numbness and tingling, ruling out other causes through extensive tests. AS was determined as the likely cause of the neuropathy. This underscores the varied clinical presentations of AS and highlights the need for further research on its potential neurological complications.

AUTHOR CONTRIBUTIONS

Amirreza Khalaji: Writing – original draft; writing – review and editing. **Susan Kolahi:** Supervision; validation; visualization. **Leyla Ghadakchi:** Data curation; investigation. **Mehdi Jafarpour:** Writing – original draft; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest to declare.

DATA AVAILABILITY STATEMENT

The data supporting the findings of this research are available upon reasonable request from the corresponding author.

ETHICS STATEMENT

This study was performed according to the principles outlined by the World Medical Association's Declaration of Helsinki on experimentation involving human subjects, as revised in 2000, and has been approved by the ethics committee of the Tabriz University of Medical Sciences.

CONSENT

Written informed consent was obtained from the patient to publish this report and clinical images. Consent has been signed and collected in accordance with the journal's patient consent policy.

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