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Case Report

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Unusual cervical involvement in psoriatic arthritis and the presence of cervical and facial dystonia: a case report

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ABSTRACT

Psoriatic arthritis (PsA) exhibits the most prevalent occurrence of cervical spine involvement within the spondyloarthropathies. The disease can progress and cause C1-C2 arthritis, erosions of the odontoid process, atlantoaxial instability, syndesmophytes, ankylosis, osteitis, ossifications, and spinal cord compression. Dystonia is a neurological movement disorder that causes involuntary and prolonged muscle contractions, twisting, repetitive movements, and atypical body positions. Cervical dystonia can predispose the atlantoaxial joint to unstable changes. Atlantoaxial degeneration has the potential to lead to cervical dystonia. This report presents a clinical case of an individual afflicted with PsA as well as cervical and facial dystonia, which includes the manifestation of atlantoaxial degeneration and pannus formation. The case was successfully managed through the administration of secukinumab and *Botulinum* toxin injections.

Keywords: Psoriatic arthritis, dystonia, *Botulinum* toxin

INTRODUCTION

Psoriatic arthritis (PsA) is a chronic inflammatory arthritis associated with psoriasis that exhibits many clinical similarities to other spondyloarthropathies and rheumatoid arthritis. This condition is classified as one of the seronegative spondyloarthropathies, as it exhibits negative serology for the rheumatoid factor and involves the axial skeleton.[1] Inflammatory symptoms include skin lesions, dactylitis, nail dystrophies, enthesitis, peripheral arthritis, and axial skeletal involvement.[2] Within the category of spondyloarthropathies, it has been observed that PsA exhibits the most prevalent occurrence of cervical spine involvement.[3] Dystonia is a neurological condition that manifests as a movement disorder characterized by the presence of involuntary and prolonged muscle contractions, resulting in twisting, repetitive movements, and atypical body positions. The occurrence of these muscular contractions can lead to atypical, frequently distressing, and sporadically incapacitating movements. In this report, we present a clinical case of cervical involvement in psoriatic arthritis, along with cervical dystonia, oromandibular dystonia, and blepharospasm.

CASE

A 55-year-old male patient presented with complaints of inability to move his neck, a right neck pulling sensation, head tilting to the right side, facial contractions, inability to eat due to contractions, and decreased quality of life. His symptoms persisted for two years. He had a medical history of diabetes mellitus, hypertension, smoking (40 packs/year), and psoriatic arthritis, but no family history of PsA or dystonia. He was diagnosed with psoriasis 7 years ago and PsA 1 year prior. Tests were negative for rheumatoid factors, anti-CCP antibodies, antinuclear antibodies, and HLA B27 antigen. Physical examination revealed facial asymmetry, with a noticeable contraction around the chin and eyes. Additionally, the right rotation of the neck and the left rotation of the chin were observed (Figure 1). The range of active cervical extension was restricted to 20 degrees, whereas left lateral flexion and rotation were limited to 30 degrees. The right sternocleidomastoid, right trapezius, and left splenius capitis exhibited hypertonicity and hypertrophy. The application of the Soto-Hall test, which involves approximating the chin to the chest, resulted in the replication of neck pain. During the neurological examination, it was observed that the pupils were of equal size (isochoric), the cranial nerve examination yielded normal results, the deep tendon reflexes were within the expected range of activity (normoactive), the cerebellar tests were performed with proficiency, and the upper and lower extremity muscle strength was found to be within normal limits. The diagnosis of dystonia was conclusively established, and the dystonic muscles were identified through the utilization of electrophysiological studies. The patient was treated with methotrexate for PsA. No active rashes were observed. However, the patients also had inflammatory back and neck pain. The results for other systemic investigations including cardiovascular, pulmoner,

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Figure 1. Image depicting the pre-treatment condition of the patient, demonstrating spasmodic torticollis, oromandibular spasm, and blepharospasm

gastrointestinal, renal systems' physical examination and laboratory test results were normal. Cranial and cervical magnetic resonance imaging (MRI), which was requested for the preliminary diagnosis of dystonia, showed sclerosis in adjacent bone structures, calcifications, and ossifications in the joint space and a 4.5x3 cm pannus formation extending to the clivus at the level of the C2 vertebral base, leading to atlantoaxial distention (Figure 2). An MRI of the sacroiliac joint performed six months previously showed marked narrowing of the inferior sacroiliac joints, increased sclerosis in the surfaces adjacent to the joint, and bone marrow edema (Figure 3). Due to the failure of other treatments, secukinumab was started. In addition, *Botulinum* toxin injections into the dystonic muscles identified by electromyography (EMG) and clinical examination were planned for symptomatic relief. Ultrasound-guided *Botulinum* toxin (BoNT) injection into the right sternocleidomastoid, right trapezius muscle, right and left semispinalis and capitis, right and left masseters, right and left temporal muscles and blind injection into the right and left orbicularis oculi, the procerus, and corrugator supercilii muscles was performed. A decrease in the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) score from 7 to 2 after the administration of the treatments showed a regression in inflammatory back and neck pain. Additionally, no active rash was observed, and both blepharospasm and oromandibular dystonia exhibited complete resolution. The hypertonicity in the sternocleidomastoid (SCM) muscle and its impact on cervical rotation have not been fully resolved. However, significant improvements were observed in terms of reduced rotation angle and alleviation of cramping (Figure 4).

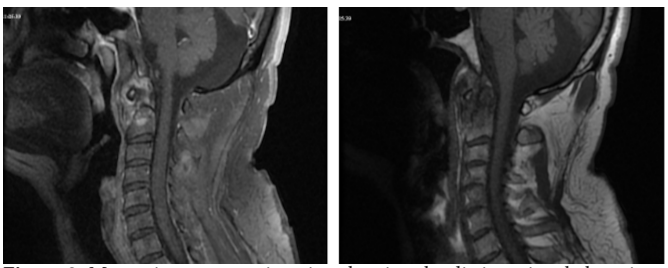

Figure 2. Magnetic resonance imaging showing the distinct signal alterations at the C2 vertebra level and atlantoaxial distention These alterations are evident in both the sagittal short tau inversion recovery and sagittal T1 fast recovery fast spin echo images

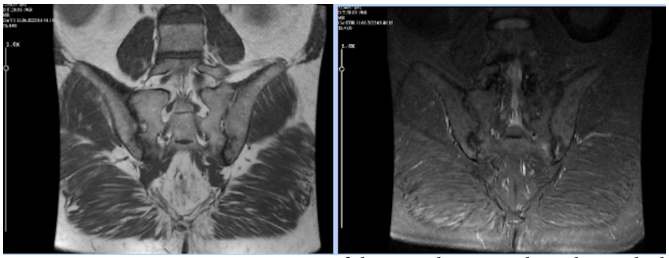

Figure 3. Magnetic resonance images of the sacroiliac joint show the marked narrowing of the inferior sacroiliac joints, increased sclerosis in the surfaces adjacent to the joint, and bone marrow edema on short tau inversion recovery and coronal T1 sections

Figure 4. Image depicting the patient clinic during the ninth month of the treatment period

DISCUSSION

PsA is likely to affect the cervical spine in the initial phase of the disease, and compared to other spondyloarthropathies, it has the highest rate of cervical involvement. The majority of patients experiencing cervical symptoms show severe and long-lasting disease.[4] C1-C2 arthritis, odontoid erosions, atlantoaxial instability, syndesmophytes, ankylosis, osteitis, ossifications, and spinal cord compression are cervical lesions that are linked to PsA.[5] Radiological involvements of the cervical spine in PsA have been reported in 35-75% of cases.[3,6,7] Lesions in the upper cervical spine are similar to the changes seen in rheumatoid arthritis, which include a periodontoid synovial pannus mass and a few cases of atlantoaxial subluxation.[3,6] We observed sclerosis, calcifications, and ossifications in the joint space in this case, where the pannus in the cervical spine extends to the clivus and widens the atlantoaxial joint. Consistent with the literature, the patient's medical history indicated a seven-year period of inflammatory back and neck pain that did not respond to treatment.

For patients with active PsA, who have not experienced relief with conventional synthetic disease-modifying antirheumatic drugs, it is recommended to switch to either a tumor necrosis factor antagonist (TNF-a), an interleukin-17 inhibitor, or an interleukin-12/23 inhibitor biologic.[8] A connection between TNF inhibitors and demyelinating diseases has been reported.[9] Additionally, there have been case reports or case series linking TNF to movement disorders, including Parkinsonism and dystonia.[10,11] Based on the current case, inhibition of IL-17 is preferred over TNF-a inhibitors.

Dystonia is a syndrome that is characterized by frequent contractions resulting in repetitive movements or abnormal postures that may be sustained or intermittent.[12] Movement patterns and postures resulting from dystonia are often highly variable and unusual, which makes it one of the most challenging movement disorders to identify. Spasms of the neck muscles or abnormal head movements occur intermittently due to contractions of the sternocleidomastoid, trapezius, and posterior cervical muscles.[12] The most frequent cause of dystonia is central nervous system pathology. However, non-neurological conditions, such as atlantoaxial subluxation and cervical degeneration, are also linked with dystonia, and they contribute to the ailment.[13] Long-term cervical dystonia was also reported to lead to craniovertebral junction narrowing, bulging, atlantoaxial joint malalignment, and myelomalacia.[14-16] A case series from 2023, including 3 patients, asserted that dystonia could lead to atlantoaxial instability through the development of the acquired os odontoideum.[17] Kawanishi et al.[18] reported that the quadriparesis of a patient with cord compression due to long-term cervical dystonia improved with repeated *Botulinum* toxin injections within 2 years. In this scenario, cervical dystonia might have worsened the atlantoaxial degeneration. Thus, the treatment methods were scheduled for both psoriatic arthritis and dystonia.

Other movement disorders, such as cranial dystonia, blepharospasm, bruxism, and burning spasms, may accompany cervical dystonia.[13] In this case, the patient had blepharospasm and oromandibular dystonia that accompanied cervical dystonia and contributed to a reduced quality of life. Primary cranial or cervical focal dystonia is commonly treated with BoNT type A.[19,20] To relieve symptoms, the patient received BoNT type A injections. Significant improvement was achieved after three seasons of BoNT injections at 3-month intervals and nine months of secukinumab treatment. As far as we know, this is the first case report of atlantoaxial involvement in PsA and its coexistence with cervical and facial dystonia.

CONCLUSION

To our knowledge, this is the first case report that describes the coexistence of PSA with dystonia. Dystonic characteristics could be explained by disease involvement in the atlantoaxial region. In this scenario, IL-17 inhibition was favored over TNF inhibition, as anti-TNF medication has been linked to movement disorders. Blepharospasm, cervical, and oromandibular dystonia were treated with BoNT type A injections. Consequently, the implementation of an appropriate treatment regimen led to a notable improvement in the quality of life for this difficult case.

ETHICAL DECLARATIONS

Informed Consent

The patient signed and free and informed consent form.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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