Ankylosing Spondylitis with Undiagnosed Concurrent Diffuse Idiopathic Skeletal Hyperostosis in a 54-year-old Male Seeking Chiropractic Care: A Case Report

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ABSTRACT

Background: Diffuse idiopathic skeletal hyperostosis and ankylosing spondylitis are conditions characterized by ossification of ligament and tendon attachments to bone, mainly impacting the axial skeleton, causing symptoms of spinal stiffness and pain. Sacroiliac joint disease is a prevalent feature of ankylosing spondylitis and has long been used as an exclusion criterion for diffuse idiopathic skeletal hyperostosis.

Case Presentation: A 54-year-old White male with a 16-year history of diagnosed but untreated ankylosing spondylitis sought care at a chiropractic clinic for chronic neck pain and spinal stiffness. Physical examination yielded findings of reduced spinal motion, hypokyphosis, and hypolordosis. Thoracic and lumbosacral radiographs revealed findings of partial sacroiliac joint ankylosis, syndesmophytes, squared vertebrae, and ossified posterior spinal ligaments. The Cervical radiographs demonstrated thick bulky ossification of the cervical anterior longitudinal ligament with minimal disc degeneration.

Results: The patient's prior diagnosis of ankylosing spondylitis was confirmed, along with a new diagnosis of concurrent cervical diffuse idiopathic skeletal hyperostosis. Treatment with spinal manipulation or with adjunctive therapies was deferred, pending rheumatology consultation.

Conclusion: Spinal stiffness, pain, and postural changes are common clinical features of

both ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis. Distinction between the two diseases is aided by evaluation of the radiographic findings. The radiographic evidence of sacroiliitis has historically excluded the diagnosis of diffuse idiopathic skeletal hyperostosis. This case report describes concurrent diagnoses of ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis that have thus far been rarely reported in scientific literature and reviews the clinical features, radiographic findings, and complications of each disease.

Key Words: Ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, HLA-B27, syndesmophytes, spondyloarthropathy

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), traditionally considered a non-inflammatory arthropathy, and ankylosing spondylitis (AS), an inflammatory spondyloarthropathy, are the two most common diseases characterized by radiographic evidence of bony proliferation, primarily involving the ligament and tendon attachments (entheses) in the axial skeleton. Both diseases are complex disorders of unknown etiology, marked by distinctive features of ectopic mineralization of soft tissues and eventual ankylosis.

First detailed by Jacques Forrestier and Juane Rotes-Querol in 1950 as 'senile ankylosing vertebral hyperostosis' and Forrestier disease, DISH was further described in younger patients and in sites beyond the axial skeleton by subsequent researchers.³ The prevalence of DISH varies significantly between populations, estimated at 2.9% in the Asian population over 50 years of age and 42% in European men older than 65 years. Higher incidences are seen in elderly and obese persons and those with hypertension and type 2 diabetes mellitus.^{4,5} The origin and development of DISH is poorly understood; possible etiologies of the disease include low Dickkopf-1(DKK1) protein levels, high vitamin A levels, and metabolic disorders, including diabetes mellitus, dyslipidemia, and hyperuricemia.^{5,6}

Ankylosing spondylitis is the most common of the seronegative axial spondyloarthropathies (axSpA), a group of disorders that includes psoriatic arthritis (PsA), inflammatory bowel disease-related arthritis (IBD), and reactive arthritis (ReA). The estimates of AS in the US population range from 0.2% to as high as 1% according to a 2009-2010 National Health and Nutrition Survey. A single population-based study of AS spanning a 30-year period from 1980 to 2009 was conducted in Olmsted County, Minnesota, revealing a 3:1 male to female prevalence in a predominately White population. In 1983, Jumshyd and Khan conducted a longitudinal study in 63 consecutive patients with AS, finding a near equal incidence between female and male patients and more frequent occurrence of the extraspinal sites of hyperostosis in Black patients. In a recent retrospective cohort study of 728,556 US military service members screened for low back pain, Nelson et al. found the incidence of AS was comparable for females and males, challenging the characterization that females are less commonly affected by the disease. The onset of AS symptoms typically manifests between the second and third decades of life. Seronegative spondyloarthropathies have a high association with the prevalence of Human Leukocyte Antigen (HLA)-B27;

approximately 2-8% of the unaffected population are *HLA-B27* positive, while 90% of patients with AS are positive for the *HLA-B27* antigen.^{7,11} The presence of *HLA-B27* is linked to sacroiliac joint osteitis and spinal entheseal osteitis (bony inflammation at tendon and ligament insertions), and the classic imaging findings of axial spondyloarthropathies, including bone marrow edema, vertebral body corner erosions (Romanus lesion) and adjacent sclerosis (shiny corner sign), and vertebral body squaring.¹² (**Table 1**)

Both DISH and AS are marked by enthesopathies of primarily the axial skeleton and are radiographically identified by ossification of ligament and tendon attachments to bone and eventual ankylosis. ^{11,13} In 1976, Resnick and Niyawama evaluated the full spine sectional radiographs of 100 patients with the spinal manifestations of DISH for the purpose of defining "strict radiographic features" as criteria for the diagnosis of DISH and its distinction from other spinal pathologies. ¹⁴ Those criteria included: "the presence of 'flowing' calcification and ossification along the anterolateral aspects of at least 4 contiguous vertebral bodies with or without associated localized pointed excrescences (bony projections) at the intervening vertebral body-disc junctions"; "a relative preservation of disc height in the involved areas and the absence of extensive radiographic changes of 'degenerative' disc disease, including vacuum phenomena and vertebral body marginal sclerosis"; and "absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis or bony fusion". ¹⁴ (**Table 1**)

The sacroiliac articulation is a complex joint, comprising a vertically oriented true synovial joint at the anterior-inferior segment and a horizontally oriented superior-posterior osseous crevice, stabilized by ligaments. Pathological changes to the sacroiliac joint can be assessed based on the site and presence or absence of characteristic radiographic features, such as erosions, sclerosis, and ankylosis. Bilateral and symmetrical sacroiliitis is a hallmark feature of ankylosing spondylitis, seen in most patients eventually diagnosed with AS. Other diseases can mimic the sacroiliitis features of AS, including Paget disease, osteitis condensans ilii (OCI), infections (septic) sacroiliitis, sarcoidosis, and other seronegative spondyloarthropathies. Subchondral sclerosis and joint space narrowing, without erosions, are classic radiographic findings of degenerative arthritis. 15

The clinical presentations of patients with *advanced* AS and DISH are similar, each exhibiting diminished spinal motion, postural changes, and spinal rigidity. ¹⁶ Distinguishing between the radiographic features of AS and DISH is seemingly uncomplicated through application of the 1976 Resnick exclusion criterion of sacroiliac joint erosions radiographs of patients with DISH. With greater availability and increased application of MRI in the diagnosis of joint disorders, significant similarities in the inflammatory characteristics of new bone formation and bone marrow edema in both inflammatory and non-inflammatory diseases have been revealed. Bone marrow edema, classically associated with the inflammatory effects of ankylosing spondylitis, may be seen with MRI in early cases of DISH, despite the disease's categorization as a non-inflammatory arthropathy. Bone marrow edema is identified by decreased (low/dark) signal on T1-weighted images and increased (high/bright) signal on T2-weighted and STIR images.^{3,11} The Resnick criterion of the absence of sacroiliac joint disease for a positive diagnosis of DISH excludes the simultaneous presence of AS.¹⁴ (**Table 1**)

We searched 8 databases with combinations of the following search terms: ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, ankylosing hyperostosis, concurrent, simultaneous, and coexistence. We filtered 383 results for case reports and case series, identifying only forty documented cases of concurrent ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis.

CASE PRESENTATION

A 54-year-old male laborer sought care as a new patient at a chiropractic clinic for chronic neck pain and spinal stiffness. His history was remarkable for non-traumatic low back pain that began at about age 19 and was first treated successfully with spinal manipulation at a different chiropractic clinic, ibuprofen, and massage. He continued care intermittently for approximately 4 years, ceasing care when he noted his symptoms worsened for several days following each treatment. At age 27, the patient initiated care in an allopathic setting for a workplace injury and was treated over the course of the following 10 years with two lumbar radiofrequency (RF) denervation procedures and ibuprofen. The patient reported his first RF denervation significantly reduced his pain for approximately 5 years; the second RF denervation worsened his symptoms. The patient was referred to a rheumatologist and was prescribed, but refused, an SSRI antidepressant. The patient, at 38 years of age, consulted a second rheumatologist and was diagnosed with ankylosing spondylitis following a positive hematology study for inflammatory markers, including HLA-B27; psoriasis, uveitis, dactylitis (swollen digits), and inflammatory bowel disease were not identified. The patient did not remember undergoing imaging evaluations, and no prior imaging was found in the local hospitals' Picture Archiving and Communication Systems (PACS). In the ensuing years following the diagnosis of AS, the patient engaged in a self-directed regimen of lengthy walking (up to 5 miles per day) and a ketogenic diet. The patient reported he lost excess weight, and his symptoms were remarkably reduced, causing him to question the accuracy of the ankylosing spondylitis diagnosis. Despite the improvements in his symptoms, the patient can work only intermittently due to pain and restricted movements. The patient chose to resume chiropractic care after viewing a Y-strap axial decompression video posted on a social media site; the video was not created, posted, or associated with the chiropractic clinic at which he was examined.

Examination

During physical examination, the physician observed the patient's limited cervical mobility, pronounced thoracic hypokyphosis and lumbar hypolordosis, and rigid posture with inability to stand fully upright. Reflex and muscle strength deficits were not detected. The patient had difficulty rising from a seated position (positive Minor sign), experienced focal pain bilaterally with cervical foraminal compression tests, and bilateral focal cervicothoracic pain with shoulder depressor tests. The patient underwent radiographic evaluation of his cervical, thoracic, and lumbosacral regions.

Imaging

Thoracic Spine Radiographs: Thin marginal bridging syndesmophytes are present from T7 to T12 (ossification of the annular/outer disc fibers, bamboo spine sign); subtle squaring of the T7-12 anterior vertebral body margins is apparent, consistent with the patient's diagnosis of AS. Disc space narrowing with osteophytosis is evident at T5 and T6. The thoracic spine is hyperkyphotic. *These findings are consistent with AS and osteoarthrosis*. (Figures 1 and 2)

Lumbar Spine Radiographs: Thin marginal bridging syndesmophytes are present from L1-S1 (bamboo spine sign); subtle squaring of the L1-L5 anterior vertebral body margins is apparent. Disc (nucleus pulposus) calcification is apparent at L1-L5. Ossification of the L1-S2 interspinous and supraspinous ligaments is seen (dagger sign); ossification of the L2-S1 zygapophyseal (facet) capsules is present (railroad track sign; railroad track sign with dagger sign is termed trolley track sign). Calcification of the bilateral iliolumbar ligaments is apparent. The joint spaces of the bilateral sacroiliac articulations are indistinct at the superior and inferior margins; minimal joint space is identified in the middle segment of the bilateral sacroiliac articulations. Zygapophyseal facet arthrosis is identified at L1/L2-L5/S1, without spondylolisthesis. Superior and axial joint space narrowing with femoral head osteophytosis is seen at the bilateral iliofemoral articulations. The lumbar spine is hypolordotic with posterior weight-bearing and a right list. *These findings are consistent with AS and osteoarthrosis*. (Figures 3 and 4)

Cervical Spine Radiographs: Dense bulky ossification of the anterior longitudinal ligament is present from C3-C6; ossification of the posterior longitudinal ligament is not apparent. Disc space narrowing with osteophytosis is present at C5 and C6; osteophytosis without appreciable disc space narrowing is evident at C2-C4. Uncinate hypertrophy is noted at C3-C7. Zygapophyseal (facet) arthrosis is identified at C2/C3-C7/T1, without spondylolisthesis. The cervical spine is hypolordotic with anterior weight-bearing. Nuchal ligament ossification is present at C5 and C6. A small round calcification is present within the left carotid artery. These findings are consistent with DISH, osteoarthrosis, and carotid arterial calcification. Findings of ankylosing spondylitis in the cervical spine are not detected. (Figures 5 and 6)

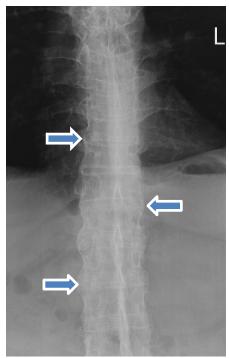


Figure 1: Anteroposterior lower thoracic/upper lumbar radiograph demonstrating multiple levels of thin bridging syndesmophytes at the right and left lateral disc margins (bamboo spine, arrows).

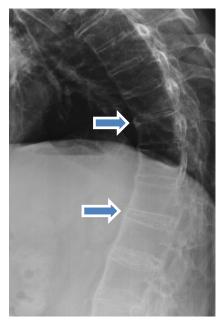


Figure 2: Lateral lower thoracic/upper lumbar radiograph demonstrating multiple levels of thin bridging syndesmophytes at anterior disc margins (arrows) and loss of the anterior concavity of the vertebral bodies (squared vertebrae).

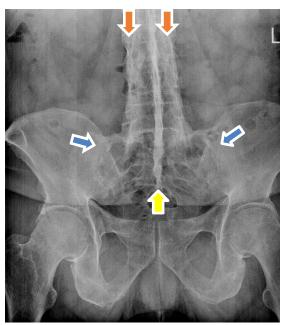


Figure 3: Anteroposterior lumbopelvic radiograph demonstrating ossification of the zygapophyseal (facet) capsules (railroad track sign, orange arrows), ossification of the supraspinous and interspinous ligaments (dagger sign, yellow arrow), and calcification of the bilateral sacroiliac ligaments with partial ankylosis of the sacroiliac joints (blue arrows).

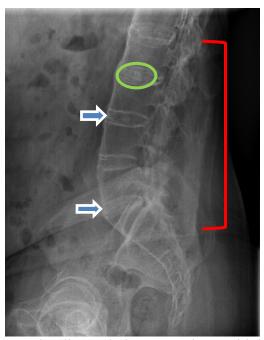


Figure 4: Lateral lumbosacral radiograph demonstrating multiple levels of thin bridging syndesmophytes at anterior disc margins (arrows), loss of the anterior concavity of the vertebral bodies (squared vertebrae), preserved disc spaces, disc (nucleus pulposus) calcifications (L1-L5, green oval), and supraspinous and interspinous ligament ossification (dagger sign on the AP view, red bracket).

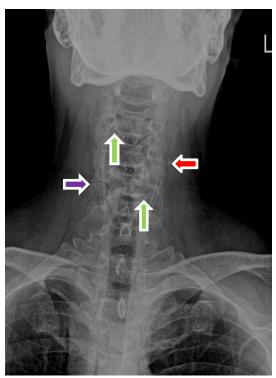


Figure 5: Anteroposterior cervical radiograph demonstrating disc space narrowing, uncinate hypertrophy (green arrows), zygapophyseal (facet) arthrosis (purple arrow), and calcification within the left carotid artery (red arrow).

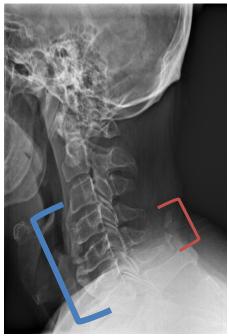


Figure 6: Lateral cervical radiograph demonstrating ossification of the anterior longitudinal ligament from C3-C7 (blue bracket), nuchal ligament ossification at C5 and C6 (orange bracket), relatively preserved disc spaces and osteophytes.

Typical Disease Features	Ankylosing Spondylitis	Diffuse Idiopathic Skeletal Hyperostosis
Age of onset	Adolescence and young adults	Over 45
Population	Males ≥ Females	Males > Females
Symptoms (dependent upon disease progression)	Early onset of back pain, reduced spinal mobility, hyperkyphosis	Asymptomatic early, reduced spinal mobility, hyperkyphosis, dysphagia
Serological Studies	+HLA-B27, +CRP, +ESR	Hyperglycemia, hyperlipidemia, hyperuricemia, excess Vitamin A
Complications	Higher risks of depression, vertebral body fractures, decreased chest expansion, cardiopulmonary compromise	Higher risks of depression, vertebral body fractures
Associated Conditions	Uveitis, inflammatory bowel disease	Diabetes mellitus, obesity, OPLL
Differential Diagnoses	Enteropathic arthritis, early psoriatic arthritis, septic arthritis, OCI	Degenerative disc disease
	Imaging Findings	Imaging Findings
	Thin annular disc fiber ossification (syndesmophytes)	Preservation of disc spaces in the early phases
	Squared vertebral bodies	Thick, bulky ossification of the anterior longitudinal ligament over 4 contiguous segments (Resnick criteria)
	Vertebral body corner erosions (Romanus lesion) and sclerosis (shiny corner sign)	
	Ossification of interspinous and supraspinous ligaments (dagger sign)	Extremity enthesal ossifications (whiskering)
	Ossification of zygapophyseal (facet) capsules (railroad sign)	Absence of zygapophyseal (facet) ankylosis
	Sacroillitis: erosions and eventual ankylosis	Absence of sacroiliac erosions
	MRI: bone marrow edema at entheses	MRI: bone marrow edema at entheses

Table 1: Comparison of typical disease features of ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis.

Treatment

The patient did not undergo any chiropractic or adjunctive therapy during the initial visit, pending additional evaluations. He was referred to his allopathic primary care physician; consultation with rheumatology was recommended. At the time of writing, the patient had not yet consulted with rheumatology and had not returned to the chiropractic clinic.

DISCUSSION

Ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis share several clinical features, including neck pain, spinal stiffness, and changes to spinal sagittal curvatures.³ The principal symptom of AS is chronic back pain, present in up to 80% of patients with the disease and typically beginning in the late teen or early adult years; uveitis and extremity arthritis are commonly seen.^{16,17} By contrast, the symptoms of DISH are uncommonly present prior to age 45 and extremity involvement is infrequent and limited to non-inflammatory enthesopathy in the shoulders and hips.¹¹ (**Table 1**)

Vertebral fractures occur 4 times more frequently in persons with ankylotic spines than in the general population, with a potential devastating consequence of spinal cord injuries. The reduced flexibility of spines with DISH and AS are less able to dissipate the energy of even seemingly minor low-velocity impacts. ¹¹ Fracture through the atypical stress points of the intervertebral disc can extend to the posterior spinal column, permitting atypical spinal

motion with a higher risk of spinal cord injury. ^{18,19} Patients with DISH and AS do not demonstrate a greater risk of peripheral fractures. ⁵

Patients with DISH may experience complications unseen in AS. The characteristic thick, bulky ossification of cervical anterior longitudinal ligament in DISH can result in dysphagia, hoarseness, and stridor from compression of the posterior pharyngeal and tracheal walls; though dysphagia is rare, it is the most common cervical symptom of DISH. ²⁰ Surgical resection of the bulky anteriorly projecting enthesophytes is effective; changes in food habits and swallowing training may help patients meet nutritional requirements with reduced risks of choking and aspiration. ²⁰ Ossification of the posterior longitudinal ligament (OPLL) is seen as linear ossification immediately posterior to the vertebral bodies, most common in the cervical spine. While OPLL lacks the flowing thickness of anterior longitudinal ligament (ALL) ossification, the space-occupying consequence of OPLL spinal canal stenosis impacting the anterior spinal cord, and early symptoms of numbness, tingling, and reduced muscle strength in the hands; as cord compression worsens, lower extremity neuropathies may result. ²⁰

Less frequently studied consequences of AS and DISH are the psychological impacts of the diseases. Worldwide, patients with AS have more frequent symptoms of depression than persons without AS; in the United States, one-third of patients with AS experience "significant" symptoms of depression, including helplessness. ²¹ In 2023, Chung et al. Published a cross-sectional study of 296 patients with axial spondyloarthritis, revealing that high levels of anxiety (13.9% of subjects) and depression (8.4% of subjects) were associated with reduced function and poorer health outcomes. The researchers theorized that functional impairment, pain, and fatigue associated with inflammatory arthritis were at the root of symptoms of psychological distress, impairing the patients' responses to treatment. ²²

Treatment of AS and DISH is complex and focused primarily on preserving mobility and reducing injury risks.²³ Pharmacological treatments include short-term use of non-steroidal anti-inflammatory drugs (NSAIDs) and systemic steroids in symptomatic patients; longterm use is discouraged to avoid associated liver, renal, and gastrointestinal side effects.²³ Disease-modifying antirheumatic drugs (DMARDS), including methotrexate and sulfasalazine, are not typically therapeutic in patients with axial disease, but may aid in reducing symptoms in patients with peripheral arthritis.²³ Tobacco smoking has been associated with an increased risk of AS disease progression; smoking cessation programs should be employed. Physical therapy and exercise programs may be helpful in reducing spinal stiffness and reducing symptoms.²³ Little research is found on the role of spinal manipulation in patients with ankylotic conditions. An unfiltered search of PubMed using search phrases "ankylosing spondylitis" and "spinal manipulation" yielded 4 articles, equally divided in discussions of complications of spinal manipulation and symptom improvement following spinal manipulation. In 2017, Cornelson et al. published a case series of the diagnosis and management of 3 patients with AS in "inactive states", each experiencing improvements in clinical, laboratory, and imaging assessments.²⁴

As dynamic thrust is considered an absolute contraindication by the Centers for Medicare and Medicaid Services (CMS) in the regions near sites of "Acute arthropathies characterized by acute inflammation and ligamentous laxity and anatomic subluxation or dislocation;

including acute rheumatoid arthritis and ankylosing spondylitis"²⁵, the paucity of research of the benefits and risks of spinal manipulation is unsurprising.

CONCLUSION

Spinal stiffness, pain, and postural changes are common clinical features of both ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis. The radiographic evidence of sacroiliitis has historically excluded the diagnosis of diffuse idiopathic skeletal hyperostosis. With greater availability and increased utilization of the advanced imaging techniques of MRI and CT, detection of overlapping features between AS and DISH, such as bone marrow edema and joint ankylosis, will improve and ultimately lead to revisions of the radiographic diagnostic criteria for DISH and axial spondyloarthropathies. Clinicians are advised to assess the clinical and radiographic findings in patients with back pain, spinal stiffness, and altered sagittal spinal curvatures, and consider simultaneous inflammatory and non-inflammatory diseases as possible etiologies.

LIMITATIONS

This is a single-patient case report. Results may not be generalizable to other individuals with similar conditions.

CONSENT

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

COMPETING INTERESTS

The authors declare no competing interests.

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