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CASE REPORT

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Occurrence of bilateral sacroiliitis and ossification of anterior spinal ligaments in the same patient

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Abstract Enthesopathy and ligament calcification may occur in both the degenerative and inflammatory types of diffuse idiopathic skeletal hyperostosis and ankylosing spondylitis. Despite their fairly frequent occurence in rheumatic disease care settings, few reports of their presence in the same patient have appeared in the literature. In this report, we present the case of a male patient with chronic low back pain, who had both ossification of anterior spinal ligaments and bilateral sacroiliac joint ankylosis, and discuss the known pathogenesis of diffuse idiopathic skeletal hyperostosis.

Key words Ankylosing spondylitis (AS) · Diffuse idiopathic skeletal hyperostosis (DISH) · Sacroiliitis

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a disease characterized by the ossification of anterior and lateral spinal ligaments. Peripheral enthesopathy may or may not accompany the disease. It is seen most often in middle-aged men. Resnick[1] defined a set of criteria based on the following radiological and pathological features. (1) The presence of flowing calcification and ossification along the anterolateral aspects of at least four contiguous vertebral bodies with or without associated localized pointed excrescences at the intervening vertebral body, i.e., the intervertebral disk junctions. (2) The relative preservation of intervertebral disk height in the vertebral segment involved, and the absence of the extensive radiographic changes found in degenerative disk disease, such as vacuum phenomena and vertebral body marginal sclerosis. (3) The absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or intraarticular osseous fusion. In other words, sacroiliitis constitutes a major exclusion criteria for DISH, while it is accepted as a unique radiological criterion for ankylosing spondylitis (AS), which is an inflammatory rheumatic disease primarily involving the axial skeleton, according to both the Rome and the modified New York criteria.[2]

While clinical complaints such as morning stiffness and inflammatory-type back pain are more prominent than the radiological features in AS, the opposite is true for DISH. We present a case of the simultaneous occurrence of bilateral sacroiliitis and ossification of anterior spinal ligaments in the same patient.

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Case report

A 58-year-old man was admitted to our facility complaining of low back pain, which had lasted for 15 years. He stated that his pain had started after a vigorous movement at that time. The intensity of the pain was the same during physical activity or at rest, and did not radiate into his legs. He had no morning stiffness or nocturnal pain. He felt paresthesias in both lower extremities, and these were more prominent on the left side. His pain was not aggrevated by Valsalva's maneuver. He had no urinary incontinence, retention, polyuria, or dysuria. He had been a smoker for 30 years. His past medical history included pneumonia in childhood, and dyspepsia for the last year. Neither he nor his family had any history of psoriasis or any other rheumatic disease. Examination of his musculoskeletal system revealed limitations of flexion and extension in the cervical spine, and of estimated forward and lateral flexions and extension of the lumbar spine. His straight leg raise was 70°/70°. He had bilateral tight hamstrings and a normal range of motions of the upper and lower extremity joints. The estimated limitation of forward flexion of his lumbar spine was thought to be due to the tight hamstrings. He had no motor or sensory loss. His tendon reflexes were normoactive symmetrically;

Fig. 1. Diffuse calcification of the anterior longitudinal ligament in the cervical spine

Fig. 2. Coarse osteophytes in the thoracic spine

plantar responses were flexor bilaterally. There were no signs of synovitis or tenderness of the sacroiliac joints. The modified lumbar Schober test was 6 cm, maximal chest expansion was 4 cm, and the finger to floor distance was 25 cm. A physical examination of his respiratory system revealed bilateral basal coarsening of the lungs. There was no abnormality in other organ systems.

Laboratory tests, including blood chemistry, complete blood count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), and urinalysis, were all normal. He was found to have a negative HLA B-27 antigen.

Radiographs showed diffuse calcification of the anterior longitudinal ligament in the cervical (Fig. 1) and thoracic spine, and coarse osteophytes in the thoracic (Fig. 2) and lumbar spine. He had no zygapophyseal joint involvement. He had ankylosis of both sacroiliac joints (Fig. 3). Computerized tomography (CT) of the sacroiliac joint was interpreted as showing chronic sacroiliitis by fusion of both sacroiliac joints (Fig. 4).

Discussion

The presence of bilateral grade 4 sacroiliitis in association with impaired lumbar spinal motion fulfills the Rome and the modified New York ankylosing spondylitis diagnostic criteria. On the other hand, back pain of an inflammatory type or synovitis must be present for a diagnosis of spondyloarthropathies according to the European Spondyloarthropathy Study Group.[3] In this patient, there were no signs or history of synovitis or inflammatory-type back pain and morning stiffness. Diffuse calcification of the anterior longitudinal ligament with indistinct clinical findings was consistent with DISH. HLA B-27 negativity favored the diagnosis of DISH, although not with exact certainty, as the patient had no history of psoriasis, which could have been the reason for spinal involvement.[4–6] However, he did have bilateral grade 4 sacroiliitis, also confirmed by CT, which is a major exclusion criterion for DISH. We therefore believe that this is a case of the simultaneous occurrence of bilateral sacroiliitis and ossification of anterior spinal ligaments in the same patient.

AS and DISH in the same patient have rarely been reported in the literature, but we found 18 such cases.[7–16] Durback et al.[17] reported the cases of eight patients with DISH who were shown by CT to have abnormalities of the sacroiliac joints. It is well known that there is a high prevalence of HLA B-27-positivity in AS. This antigen was also found in DISH in one study,[18] but its prevalence was not as high as in AS. Although not confirmed by other authors,[19–21] this may indicate that these two diseases might have a common or similar genetic base. In fact, the etiology of DISH remains unclear. Local growth-stimulating factors were claimed to be initiating factors for DISH development in some studies.[22,23] DISH has also been considered to be a multisystemic hormonal disorder,[24] as disturbances of insulin, IGF-1, and growth hormone were reported in many

Fig. 3. Bilateral sacroiliac joint ankylosis

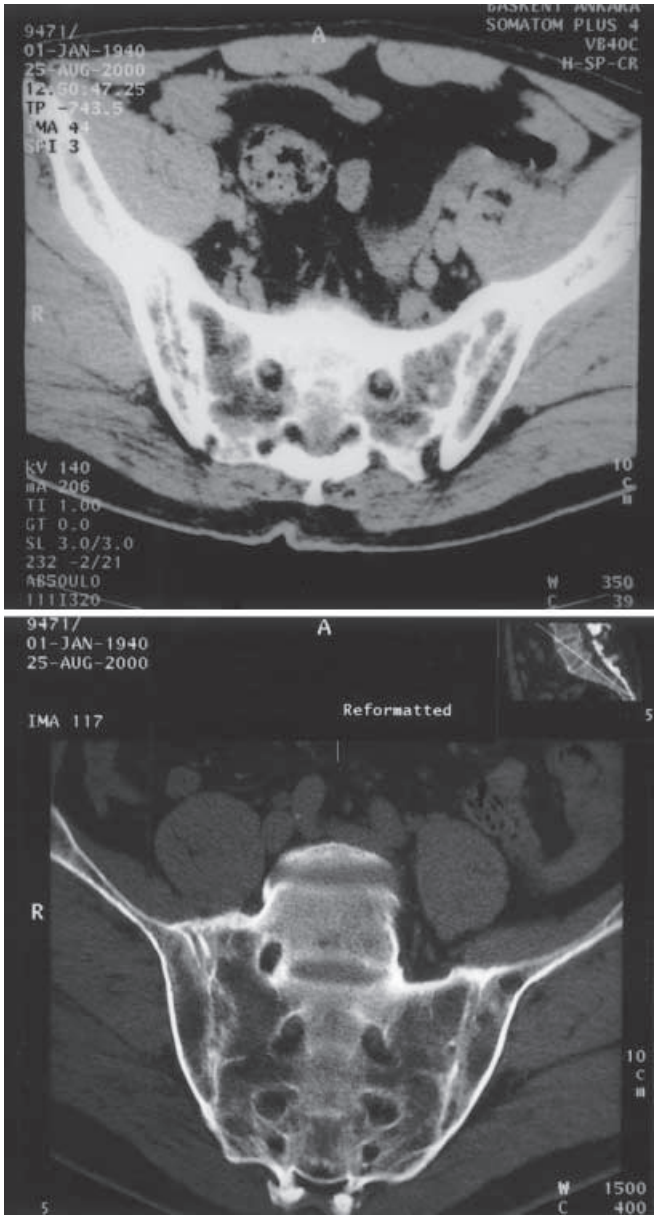

Fig. 4. Computed tomography showing chronic sacroiliitis of both sacroiliac joints

patients. El Miedany et al.[25] stated that DISH is a diffuse systemic condition which is most probably related to abnormal bone cell growth, and activity reflecting the influence of metabolic factors that lead to new bone deposition. He suggested that the vertebral blood supply is a predisposing factor that contributes to the onset, progression, and/or localization of DISH. However, which factor initiates the ossification process remains in question. Abnormal osteoblastic growth or activity in the bony ligamentous region could be the reason for new bone formation, but this poses the question, "Why do osteoblasts show abnormal growth?" The process of osteogenesis has been reported to be promoted and maintained by several growth factors, including insulin-like growth factor-1 (IGF-1) and transforming growth factor-beta (TGF-β). Patients with DISH had been reported to have elevated levels of insulin and growth hormone, which could explain the osteoblastic growth or proliferation.[24] Active TGF-β has three major biological effects: growth inhibition, stimulation of extracellular matrix formation, and immunosuppression.[26] TGF-β is an important factor in enchondral ossification and osteophyte formation.[27–29] It was found that the concentration of this inflammatory mediator determines the formation of bone and cartilage.[29] It has been clearly demonstrated that TGF-β injected in the periosteal lining of rats enhances bone formation and contributes to fracture healing.[30] The genotype of TGF-β is important in new bone formation. An association of TGF-β-1 with spinal osteophytosis in Japanese women has been studied, and it was found that the T29→C polymorphism of the TGF-β-1 gene exhibited inverse patterns of association with genetic susceptibility to spinal osteophytosis and with osteoporosis.[31] An explanation for the formation of significantly different types of osteophytes, which are coarse in spinal osteophytosis and tiny in osteoporosis, could be the involvement of zygapophyseal joints at that level and the concentration of TGF-β. Prominent zygapophyseal joint involvement in AS causes immobilization of that segment. In contrast, as there is no zygapophyseal joint synovitis in DISH, the segment is more mobile, and the greater mobility of the spine induces greater tensile forces at the outer anulus fibrosus, leading to a more pronounced local spreading of inflammation and inflammatory mediators such as TGF-β. The continuous disruption of healing tissue results in more extened healing processes, with the formation of large, plump osteophytes.

A more extensive study of the zygapophyseal joints in both AS and DISH could help the clinician to determine more easily whether the osteophytes are due to AS or DISH.[32]

Although these are not rare disorders and could share a common genetic base, they are not very often noted in the same patient. An explanation for this could be that as DISH is of relatively low clinical and therapeutic importance, it is possible that it is not reported by most physicians. Another explanation is that there may be no association between the two at all, and a simultaneous occurrence may just be coincidental. In conclusion, we made a diagnosis of simultaneous occurrence of bilateral sacroiliitis and ossification of anterior spinal ligaments in the same patient, based on his history, a physical examination, and roentgenological findings, because the findings did not fit either AS or DISH alone.

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