

Severe hip joint motion restriction in a patient with diffuse idiopathic skeletal hyperostosis

A case report

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Abstract

Rationale: Severe limitation of motion (LOM) in the spine occasionally occurs in patients with diffuse idiopathic skeletal hyperostosis (DISH). However, in extraspinal areas, significant LOM has rarely been reported. In this study, we report a patient with DISH who had severe motion restriction within both hip joints.

Patient concerns: A 57-year-old man presented with a 10-year history of LOM of bilateral hip joints. He had gait difficulty due to shortened stride length induced by LOM. Also, he had mild bilateral hip pain [numeric rating scale (NRS): 2].

Diagnoses: Hip joint range of motion was 60.3% of normal. The patient had bridging ossification along the anterolateral borders of 7 contiguous vertebrae (T10 to L4) without the findings of ankylosing spondylitis or degenerative disc disease. In addition, severe hyperostosis was diffusely formed around bilateral hip joints. He was diagnosed with DISH.

Interventions: No specific treatment was performed for controlling LOM of bilateral hip joints. Meloxicam 15 mg was administered to the patient for the management of mild bilateral hip pain.

Outcomes: At 2-month follow-up visit, the degree of LOM in the bilateral hip joints was not changed. However, the patient's pain severity was reduced from NRS 2 to 1.

Lessons: We showed that DISH can cause significant motion restriction due to severe hyperostosis in the bilateral hip joints.

Abbreviations: DISH = diffuse idiopathic skeletal hyperostosis, LOM = limitation of motion, NRS = numeric rating scale, ROM = range of motion.

Keywords: diffuse idiopathic skeletal hyperostosis, hip joint, limitation of motion

1. Introduction

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier disease, is a noninflammatory disease that leads to ossification and calcification of ligaments and entheses mainly in the spine.^[1,2] In addition to the spine, DISH can spread to any extraspinal joint in the body.^[3] DISH usually occurs in persons over age 50, with a prevalence of about 15% in women and 25%

in men over age 50.^[4] Although patients with DISH are often asymptomatic, some develop symptoms, such as pain, stiffness, and limitation of motion (LOM), in the axial or peripheral joints.^[3] The degree of LOM is usually mild.^[5] However, severe LOM can occasionally present in the spine, and markedly limited spinal mobility can limit activities of daily living and decrease the quality of life.^[6] However, severe LOM has rarely been reported in extraspinal areas.

This study describes a patient with DISH and severe motion restriction in both hip joints.

2. Case report

A 57-year-old man presented with a 10-year history of slowly progressive, restricted bilateral hip joint motion. He could independently walk without assistance or device but had gait difficulty due to shortened stride length induced by LOM in the bilateral hip joints. He also had mild bilateral hip pain [numeric rating scale (NRS): 2], but no motor weakness or sensory deficit. Upper and lower extremity deep tendon reflexes were normal, but ankle clonus and a Babinski sign were not assessed. The passive hip joint range of motion (ROM) was evaluated in supine and side-lying position using a hand-held goniometer. The bilateral ROM was as follows: hip flexion 90°, extension 0°, abduction 5°, adduction 10°, internal rotation 0°, and external rotation 20°. Compared with normal values,^[7] (flexion 120°, extension 30°, abduction 45°, adduction 30°, internal rotation 45°, external rotation 45°), hip joint ROM was 60.3% of normal. In contrast, LOM was not observed in the spine. The patient provided

Editor: N/A.

Funding/support: This work was supported by the National Research Foundation of Korea (NRF) grant funded by the Korea government (MSIT) (NRF-2017R1C1B5017714).

The authors have no conflicts of interest.

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Medicine (2018) 97:18(e0652)

Received: 26 January 2018 / Accepted: 12 April 2018

<http://dx.doi.org/10.1097/MD.00000000000010652>

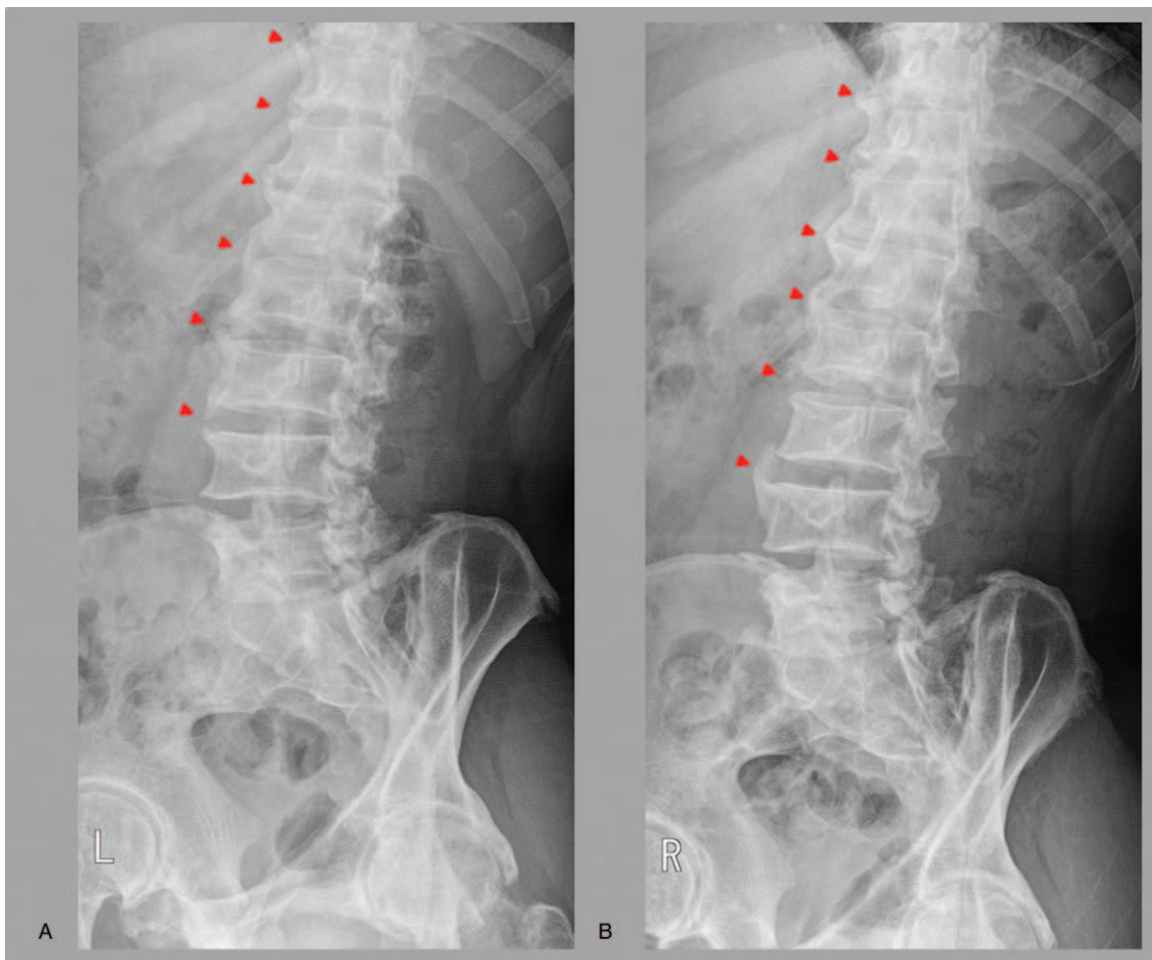


Figure 1. Left (A) and right (B) oblique thoracolumbar spine X-rays show bridging ossification of both right- and left-side anterolateral aspects of 7 contiguous vertebrae (T10 to L4) (red arrow heads).

informed consent for participation in the study. The study was approved by the Institutional Review Board of Yeungnam university hospital.

The while blood cell count, C-reactive protein, calcium, and parathyroid hormone levels were within normal range. HLA-B27 was negative. The erythrocyte sedimentation rate was elevated to 67 mm/h (normal value: <20 mm/h). The spine X-ray (Fig. 1) showed bridging ossification along both right- and left-side anterolateral borders of 7 contiguous vertebrae (T10 to L4). The disc heights in the involved spinal areas were preserved. Anterolateral bridging ossification was also observed between the T7 and T8 vertebrae. However, radiological findings of degenerative disc disease, including disc-space narrowing, vacuum phenomena, and vertebral body marginal sclerosis were not present. Pelvic X-ray and computed tomography (Fig. 2) showed diffuse, severe hyperostosis around the bilateral hip joints. The hip joint spaces were relatively well-preserved. The pelvic X-ray also showed enthesophytes on the bilateral iliac crests and ischial tubercles. Erosions, sclerosis, and intra-articular body fusion were not observed in the sacroiliac joint. According to diagnostic criteria defined by Resnick and Niwayama,^[1] we diagnosed the patient with DISH. For controlling bilateral hip pain, meloxicam 15 mg was administered to the patient. At the 2-month follow-up visit, the patient's pain severity was reduced from NRS 2 to 1. The degree of LOM in the bilateral hip joints was not changed during 2 months follow-up.

3. Discussion

The current study describes a patient with severe bilateral hip joint LOM due to severe hyperostosis induced by DISH. The LOM was symmetric and approximately 60% of normal.

DISH is a systemic noninflammatory disease characterized by ossification of the bony attachments of tendons, ligaments, and joint capsules.^[1,2] The most common site of ossification is the anterior longitudinal ligament of the spine.^[1] This ossification process was reported to be associated with the abnormal growth and function of osteoblasts in the osteoligamentary binding.^[8] Although the fundamental cause has not been clearly elucidated, genetic and external factors, including mechanical factors, drugs (isotretinoin, tretinoin, acitretin, and other vitamin A derivatives), and metabolic conditions are hypothesized to be associated with the development of DISH.^[9]

Previous studies reported that patients with DISH occasionally have marked reduction in spinal mobility as well as postural abnormalities.^[6] Accordingly, DISH can be confused with ankylosing spondylitis (AS).^[10] In our case, reduction of spinal mobility was not observed. Moreover, as our patient did not show associated radiological findings, including syndesmophytes, spondylodiscitis, apophyseal joint sclerosis and ankylosis, and sacroiliitis, AS was ruled out. In addition, our patient showed ossification along the anterolateral aspects of more than 4 contiguous vertebral bodies, with preservation of disc height in

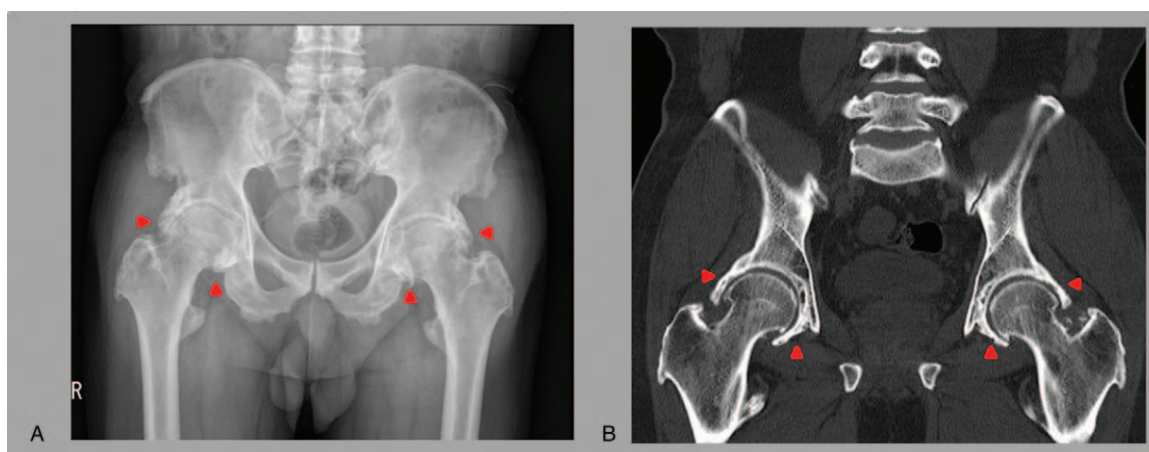


Figure 2. On anteroposterior pelvic X-ray (A) and coronal pelvic computed tomography (B), severe and diffuse hyperostosis was observed around the bilateral hip joints (red arrow heads).

the involved areas and the absence of degenerative disc disease; thus, we diagnosed DISH. In contrast to the spine, marked motion restriction in extraspinal joints has not been reported previously. However, unlike the usual manifestations of DISH, our patient had severe motion restriction in the bilateral hip joints, rather than in the spine. The marked hip joint LOM caused gait difficulty.

Fahrer et al^[11] recruited 32 patients with DISH and compared passive hip joint motion with that of 35 controls. They reported that greater juxta-hip joint ossification was found in DISH patients than in controls. However, most of these ossifications were asymptomatic and did not affect ROM. Compared with controls, the occurrence and degree of hip joint motion limitation in DISH were not significantly different. Moreover, even when patients had hip joint LOM, the authors did not provide specific data on severity. Therefore, we cannot know the extent to which DISH affected hip joint motion. To the best of our knowledge, this is the first report to show severe motion restriction in extraspinal joints in a patient with DISH.

In conclusion, we report a patient with DISH with severe hyperostosis around the hip joints and motion restriction in the joints. The bilateral hip ROM was about 60% of normal, leading to shortened stride length and impaired gait function. Unlike previous reports, we showed that DISH can cause significant motion restriction due to severe bilateral hip joint hyperostosis. However, this was a case report, and further studies involving larger numbers are warranted for elucidation of this topic.

Author contributions

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Methodology: Won Ho Lee.

Supervision: Min Cheol Chang.

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