

Sacroiliac joint involvement in ochronotic arthropathy

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A 50-year-old man was referred to our rheumatology unit for a possible diagnosis of axial spondyloarthritis (axSpA). He had chronic mechanical back pain for nearly 20 years, and his medical inquiry was negative for SpA-related features such as uveitis. Physical examination revealed restriction of spinal movements in the lumbar and cervical region and black pigmentation of the sclera and ear cartilage. X-ray films of the sacroiliac joints (SIJs) were suggestive of unilateral sacroiliitis graded as three on the left side. Thoracic and lumbar spine radiographs showed intervertebral disc calcification and vacuum phenomenon at multiple levels (Figure 1a). Computed tomography of the SIJs showed joint space narrowing, irregularity, and erosions on the left SIJ (Figure 1b). Magnetic resonance imaging (MRI) showed hypointense (Figure 1c; T1 weighted) and hyperintense (Figure 1d; short-tau inversion recovery) lesions located on the caudal iliac side of the left SIJ. Minimal contrast enhancement was observed in that location (Figure 1e; post gadolinium). Laboratory examinations showed that C-reactive protein level and erythrocyte sedimentation rate were within the normal ranges, urinary homogentisic acid (HGA) excretion was elevated (1,203 mmol/mol creatinine), and plasma tyrosine levels were normal. HLA-B27 test was negative. The patient was diagnosed with ochronotic arthropathy and treated conservatively.

Ochronotic arthropathy refers to the deposition of HGA crystals in the musculoskeletal system. Similar to SpA, spine and large joint are the most commonly affected sites in this condition (1). In general, sparing of SIJs used to be one of the main differentiating features of ochronotic arthropathy from ankylosing spondylitis. However, recent case studies reported structural SIJ changes by conventional X-ray films (2). Interestingly, these deposits may show inflammatory signal changes (3) or gadolinium enhancement on the MRI, as presented in this case. Therefore, physicians should consider SIJ involvement as one of the skeletal manifestations of ochronosis, and the presence of this feature is not sufficient to exclude ochronotic arthropathy from SpA. Flattened and calcified intervertebral discs in the radiographs along with

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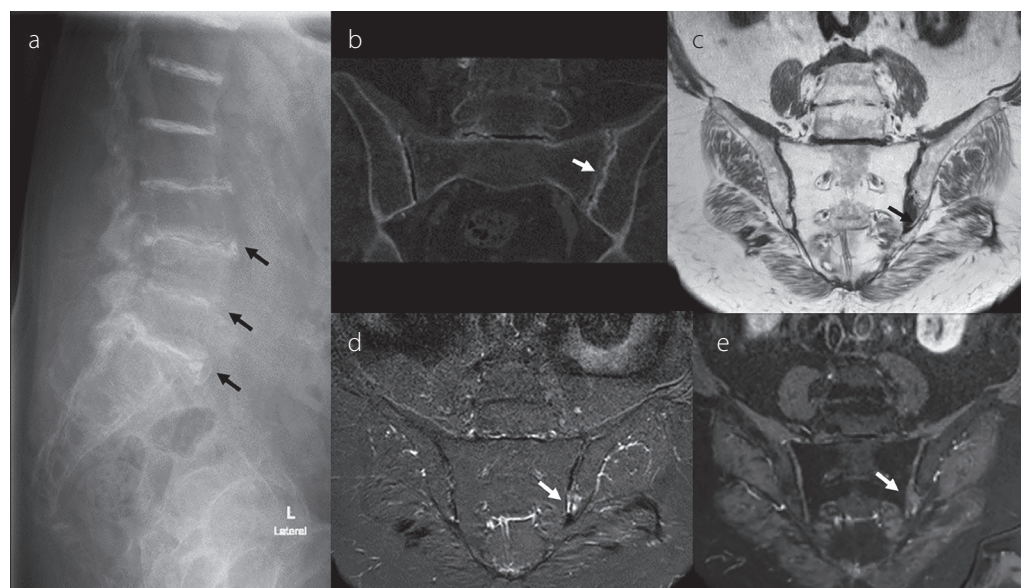


Figure 1. a-e. Radiograph of lumbar spine, showing intervertebral disc calcification and vacuum phenomenon (black arrow) (a). Computed tomography scan of the sacroiliac joints (SIJs) documented joint space narrowing, irregularity, and erosions on the left SIJ (white arrow) (b). Magnetic resonance imaging showed hypointense (c); (T1 weighted) (black arrow) and a hyperintense (d); (STIR) (white arrow) lesions located on the caudal iliac side of the left SIJ. Minimal contrast enhancement was observed in that location (e); (post gadolinium) (white arrow).

appropriate clinical symptoms such as dark urine remain the main differentiating features of ochronotic arthropathy from SpA.

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