RMD Open
Rheumatic & Musculoskeletal Diseases

LETTER

Whipple's disease: a rare cause of sacroiliitis

Arthur Bouchut ,[1] Omar Al Tabaa,[1] Elise Descamps,[1] Xavier Puechal ,[2] Christian Roux [1,3]

To cite: Bouchut A, Al Tabaa O, Descamps E, *et al*. Whipple's disease: a rare cause of sacroiliitis. *RMD Open* 2022;**8**:e002732. doi:10.1136/rmdopen-2022-002732

Accepted 21 October 2022

[1]Rheumatology, Hospital Cochin, Paris, France
[2]Internal Medicine, National Referral Center for Rare Systemic Autoimmune Diseases, Hospital Cochin, Paris, France
[3]INSERM U1153, Paris Descartes University, Reference Center for Genetic Bone Diseases - Department of Rheumatology, Cochin Hospital, Paris, France

Correspondence to
Arthur Bouchut;
arthur.bouchut@aphp.fr

Dear editor,

Whipple's disease (WD) can be diagnosed in patients with destructive arthritis.[1–3] We report a case of sacroiliitis (SI), which is unfrequently reported in WD.

A 45-year-old patient had asymmetrical inflammatory arthralgias of the wrists, hands, hips and knees, with intermittent joint swelling disappearing spontaneously within 24–48 hours, and inflammatory low back pain. He had no other significant medical history except for peptic esophagitis. There were no extra-articular symptoms. Acute phase reactants were elevated with a C reactive protein (CRP) level between 32 and 86 mg/L. Autoimmune tests and HLA B27 typing were negative. No synovial fluid was obtained. X-ray imaging showed bilateral carpal destruction, right hip destruction and bilateral SI. A diagnosis of axial and peripheral, destructive, HLA B27-negative spondyloarthritis was made. The patient was treated with nonsteroidal anti-inflammatory drugs and methotrexate, without clinical or biological improvement. Right hip destruction worsened after 1 year of treatment. Subsequent introduction of four successive tumour necrosis factor-α inhibitors (TNFi) failed to improve the patient's condition. At 59, he was referred to our rheumatology department for a 'refractory spondyloarthritis'.

When the patient was referred, he also had lost 12 kg over the 5 last years. He had dyscalculia. Acute phase reactants were elevated (CRP 93 mg/L). Joint ultrasound revealed synovitis of both wrists with Doppler activity. MRI revealed active right SI (figure 1). Full-body positron emission tomography revealed no alternative cause for weight loss or elevation of acute phase reactants. Digestive endoscopies revealed no evidence for neoplasia or an inflammatory intestinal disease. Because of the intermittent inflammatory joint involvement, weight loss, marked elevation of acute phase reactants and resistance to TNFi, WD was considered a possible diagnosis. Tropheryma whipplei (Tw) PCR assay was positive on two stool and saliva samples. To differentiate Tw colonisation from WD, Tw PCR assay was also performed on a synovial biopsy of the wrist, a duodenal biopsy and in the cerebrospinal fluid, and was positive on

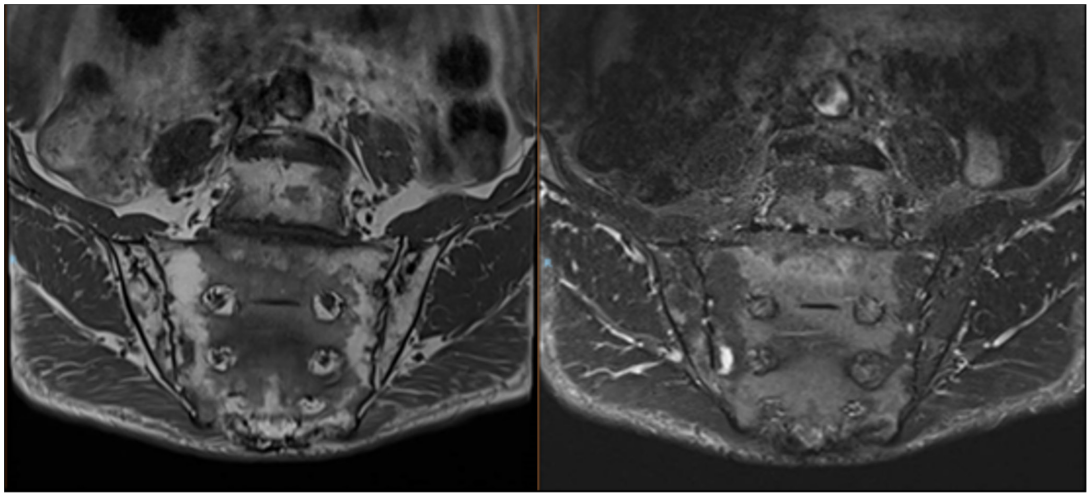

Figure 1 Right sacroiliitis on T1-weighted (left) and T2-weighted (right) MRI before treatment of WD. WD, Whipple's disease.

all samples. The final diagnosis was WD with articular, digestive and neurological involvement.

Treatment with doxycycline, hydroxychloroquine and sulfadiazine was introduced. After 9 months of treatment, the patient no longer had any tender or swollen joint. His weight was stable. Acute phase reactants were no more elevated. In contrast with clinical improvement, MRI aspects were unchanged. Joint ultrasound did not reveal any synovitis. There was no structural evolution on X-ray. The patient was considered in remission.

SI is unfrequently reported in WD. To our knowledge, 14 cases of radiographic SI are reported in patients with WD, at least 10 of which were HLA B27 negative.[4 5] Rheumatologists must be aware of WD when spondyloarthritis has intermittent inflammatory joint involvement, marked elevation of acute phase reactants and is refractory to treatment. There was no major clinical worsening under TNFi, which was consistent with other case reports.[6] Antibiotic therapy allows a significant improvement in these patients.

Contributors AB, OAT, ED, XP and CR managed the case. AB and OAT wrote the paper. AB, OAT, ED, XP and CR reviewed and approved the final manuscript.

Funding The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

Competing interests None declared.

Patient consent for publication Consent obtained directly from patient(s).

Provenance and peer review Not commissioned; externally peer reviewed.

ORCID iDs
Arthur Bouchut http://orcid.org/0000-0003-1748-2555
Xavier Puechal http://orcid.org/0000-0003-3573-9203
Christian Roux http://orcid.org/0000-0002-5880-2933

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