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IMAGES IN RHEUMATOLOGY

Vanishing hips: unveiling Gorham-stout syndrome

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ABSTRACT

Background: Gorham-Stout syndrome (GSS) is a rare disorder characterized by progressive osteolysis of unclear aetiology. It can affect various bone sites, with variable clinical presentations, being bilateral hip involvement particularly uncommon.

Case Presentation: We report the case of a 54-year-old woman with morbid obesity and type 2 diabetes, with hip and knee osteoarthritis. For over two years, she experienced progressive bilateral hip pain and gait limitation. Plain radiographs revealed bilateral resorption of the femoral heads, absent in previous radiograms. Computed tomography showed severe acetabular dysplasia with deformity and resorption of both femoral heads. Magnetic resonance imaging confirmed bone loss and bone marrow infarction. There were no clinical or analytical features suggestive of inflammatory arthropathy, nor phospho-calcium metabolism disorder apart from vitamin D deficiency. Based on the radiological and clinical findings, a diagnosis of Gorham-Stout syndrome was considered.

Conclusion: This case illustrates a rare and unusual presentation of GSS with bilateral hip involvement. Due to its rarity and non-specific clinical features, GSS is often a diagnosis of exclusion. Reporting such cases is essential to increasing awareness of this rare condition.

Keywords: Hip; Diagnostic imaging; Osteoarthritis; CT scanning; Radiology.

INTRODUCTION

Gorham-Stout syndrome (GSS), also known as vanishing bone disease, is a rare disorder characterized by progressive osteolysis of unclear aetiology[1-2]. We report a case of bilateral hip reabsorption, suggestive of GSS.

CLINICAL CASE

A 54 years old woman with morbid obesity and type 2 diabetes, with no history of smoking, alcohol consumption, or other risk factors, was referred to our consultation from Orthopaedics Department due to radiographic abnormalities.

The patient reported chronic arthralgia, predominantly in knees and hip joints, attributed to osteoarthritis, for which she was had been taking non-steroidal anti-inflammatory drugs and opioids for pain management. She described worsening of the bilateral hip pain for the past two years, with progressive gait limitation.

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Hip plain radiographies (Figure 1A) showed bilateral resorption of the femoral heads, not present in previous radiograms from 2018.

A computer tomography (CT) scan (Figure 1B) was performed, indicating severe acetabular dysplasia, with deformity and resorption of the femoral heads, particularly on the left side, and significant sclerosis on the right, with intra-articular thickening and reactive synovial thickening on the right side. T1-weighted axial (Figure C) and STIR coronal MRI sequences of the pelvis (Figure D) demonstrate bilateral absence of the femoral heads with symmetric acetabular remodelling; no associated fluid collections are seen. An incidental subtrochanteric bone infarct of the right femur is also observed.

There were no signs or symptoms suggestive of inflammatory arthropathy. Apart from vitamin D deficiency, phospho-calcium metabolism was unremarkable (normal calcium, phosphate and parathyroid hormone levels).

Although the presence of risk factors for avascular osteonecrosis, this condition usually presents with subarticular bone marrow signal intensity changes of the femoral head, which in advanced stages may result in subchondral collapse of the weight-bearing region. In the present case, the absence of these characteristic findings, together with the presence of bilateral diffuse resorption and osteolysis of the femoral heads, is more consistent with GSS.

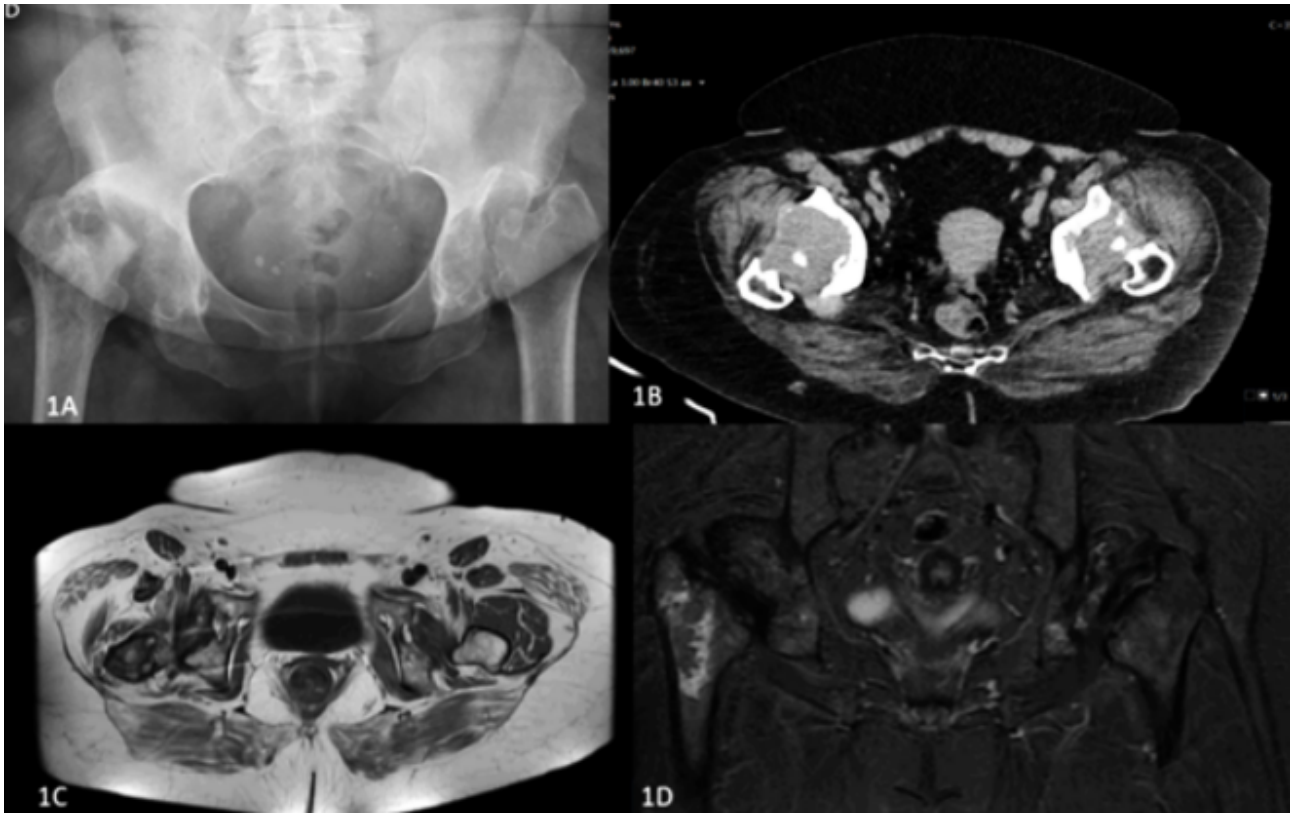

Figure 1. 1A, Anteroposterior plain radiographs of the hips showing bilateral reabsorption of the femoral heads; 1B, CT scan demonstrating severe acetabular dysplasia with deformity and resorption of the femoral heads; 1C-D, MRI scans (coronal T1-weighted (C) and STIR sequences (D) depicting bone loss and osteolysis in both femoral heads.

DISCUSSION

GSS is a rare disorder, characterized by destruction of osseous matrix and proliferation of vascular structures, leading to osteolysis[1]. It may involve one or multiple bone sites[2]. Its aetiology remains unclear and clinical presentation can vary widely depending on the affected bone[1-2]. Prognosis is variable and may depend on the presence of additional manifestations, such as pleural effusion, or the development of complications, including infection or spinal cord involvement[1].

Despite this patient having risk factors for avascular necrosis (AVN), the absence of subarticular bone marrow signal changes and subchondral collapse, together with bilateral diffuse resorption and osteolysis of the femoral heads, support the diagnosis of GSS. Clinical and laboratory evaluation excluded infectious, metabolic, endocrine and inflammatory causes. While malignancy cannot be completely ruled out without biopsy, the bilateral symmetrical involvement and the absence of constitutional symptoms or aggressive radiological features make this diagnosis unlikely.

Reporting these cases and their imaging findings is essential for enhance understanding and recognition of this rare condition.

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