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Case Report

Hypertrophic Osteoarthropathy With Acro-osteolysis in a Patient With Primary Pulmonary Hypertension☆

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ABSTRACT

Hypertrophic osteoarthropathy is an entity characterized by a triad of periostitis of long bones, clubbing and arthritis. Radiologically there are two patterns, one characterized by new bone formation which predominates in patients with pulmonary disease, and another by acro-osteolysis that is most frequently associated with congenital heart disease. We report the case of a 30-year-old man diagnosed with primary pulmonary hypertension for two years, developing hypertrophic osteoarthropathy with a mixed radiological pattern.

Osteoartropatía hipertrófica con acro-osteolisis y neoformación ósea en un paciente con hipertensión pulmonar primaria

Palabras clave:
Osteoartropatía hipertrófica
Acro-osteolisis
Neoformación ósea
Hipertensión pulmonar primaria

RESUMEN

La osteoartropatía hipertrófica es una entidad caracterizada por la tríada de periostitis de huesos largos, acropaquias y artritis. Radiológicamente se distinguen 2 patrones; uno caracterizado por neoformación ósea que predomina en pacientes con patología pulmonar, y otro por acro-osteolisis que se asocia más frecuentemente con cardiopatías congénitas. Presentamos el caso de un varón de 30 años diagnosticado de hipertensión arterial pulmonar primaria desde los 2 años, que desarrolló una osteoartropatía hipertrófica con un patrón radiológico mixto.

Introduction

Hypertrophic osteopathy (HOA) is a disease characterized by chronic proliferative periostitis of long bones, acropachy and arthritis.[1,2] It can be divided according to its etiology in primary HOA, with no apparent underlying cause, familial aggregation and chronic progression; and secondary, associated with lung, cardiac, hepatic or intestinal diseases, with a tendency to bilateralism, symmetric and rapidly progressive. There are two distinct radiological patterns of HOA, one characterized by hypertrophia or bone neoformation, predominant in patients with lung disease (pneumic HOA) and which has its onset after puberty; and another with acroosteolysis, associated frequently with cyanotic congenital heart disease and which has its onset during childhood. Two variants have been seen in this last pattern, one in which the reabsorption of the distal phalanges makes them adopt a pyramidal form and another in which its massive destruction leads to the formation of flat surfaces.[1,3] We present the case of a patient with primary pulmonary hypertension who developed HOA with a mixed radiological pattern.

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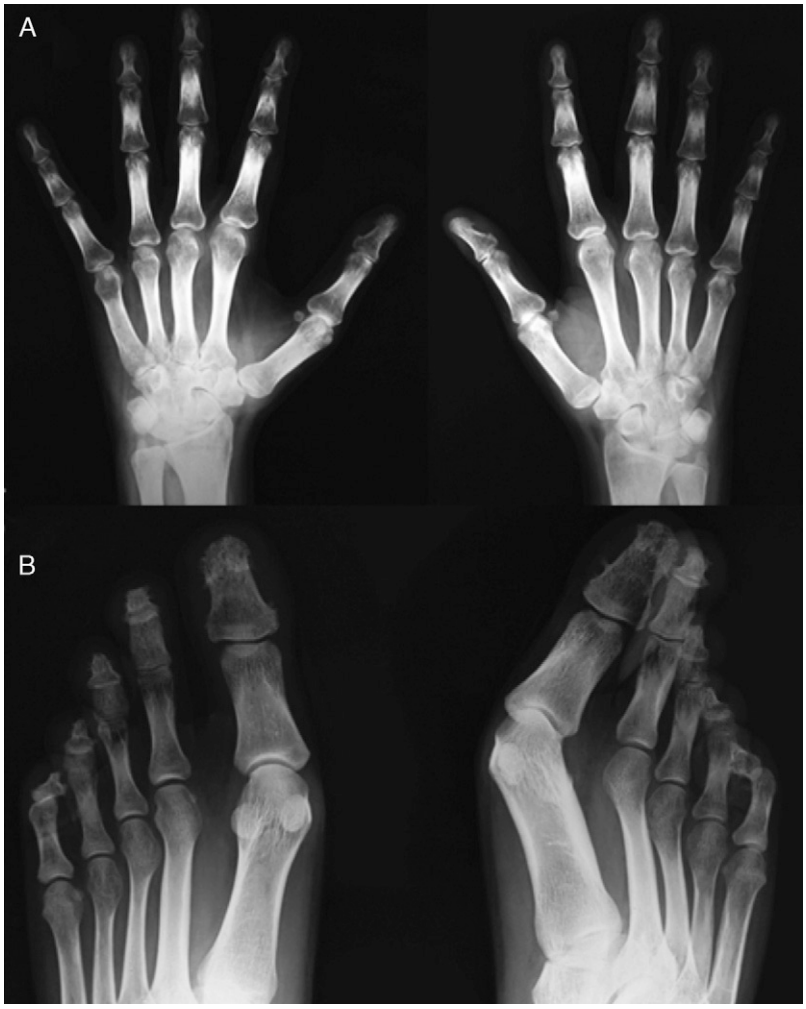

Fig. 1. Hand X-ray (A) in which there is a periosteal reaction on the radius. There is distal widening with hypertrophic changes and areas of osteolysis, with a good example on the right fourth finger. Juxtaarticular demineralization. Feet X-ray (B) observing marked destruction of the distal phalanges, some with flattened morphology and distal hypertrophy with bone proliferation. There is marked deformity of the fingers.

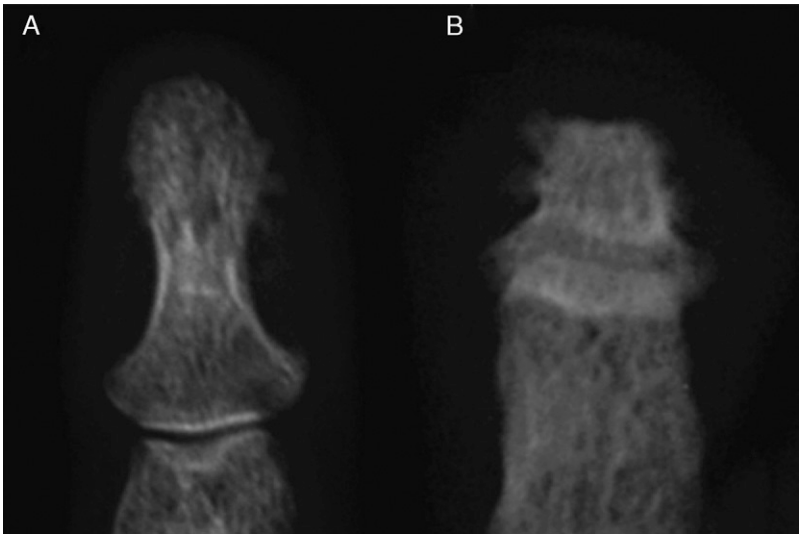

Fig. 2. Comparative image of the distal phalange of the hand (A) and foot (B); the first has proliferative changes and the second destructive changes with flattened morphology.