A rare differetial diagnosis of Paget's disease

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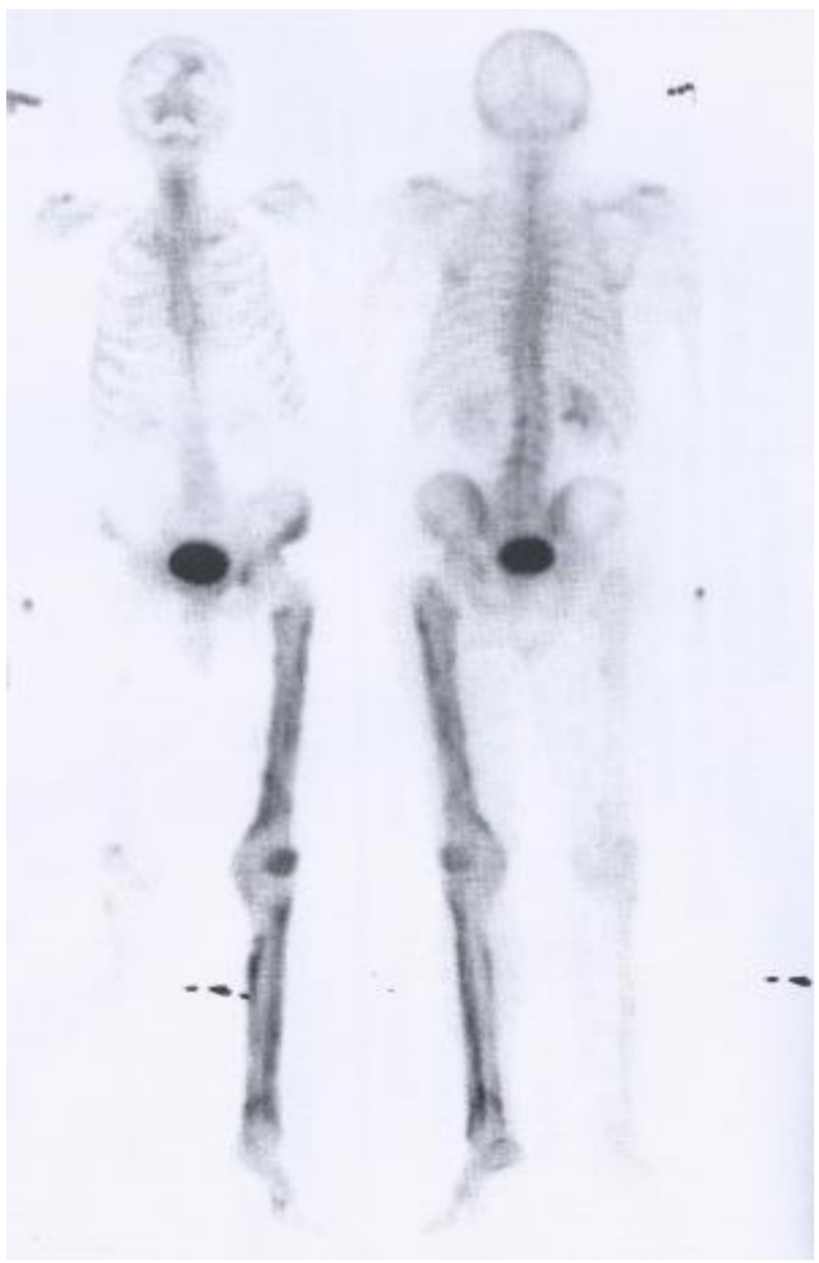
OBJECTIVES

Hypertrophic osteoarthropathy (HOA) is a syndrome characterized with proliferation of bones and skin at the distal parts of extremities. Clubbing, periostitis of tubular bones and non-inflammatory arthritis of lower extremities are commonly seen as a part of this syndrome. Here we report a case of HOA presented as Paget disease.



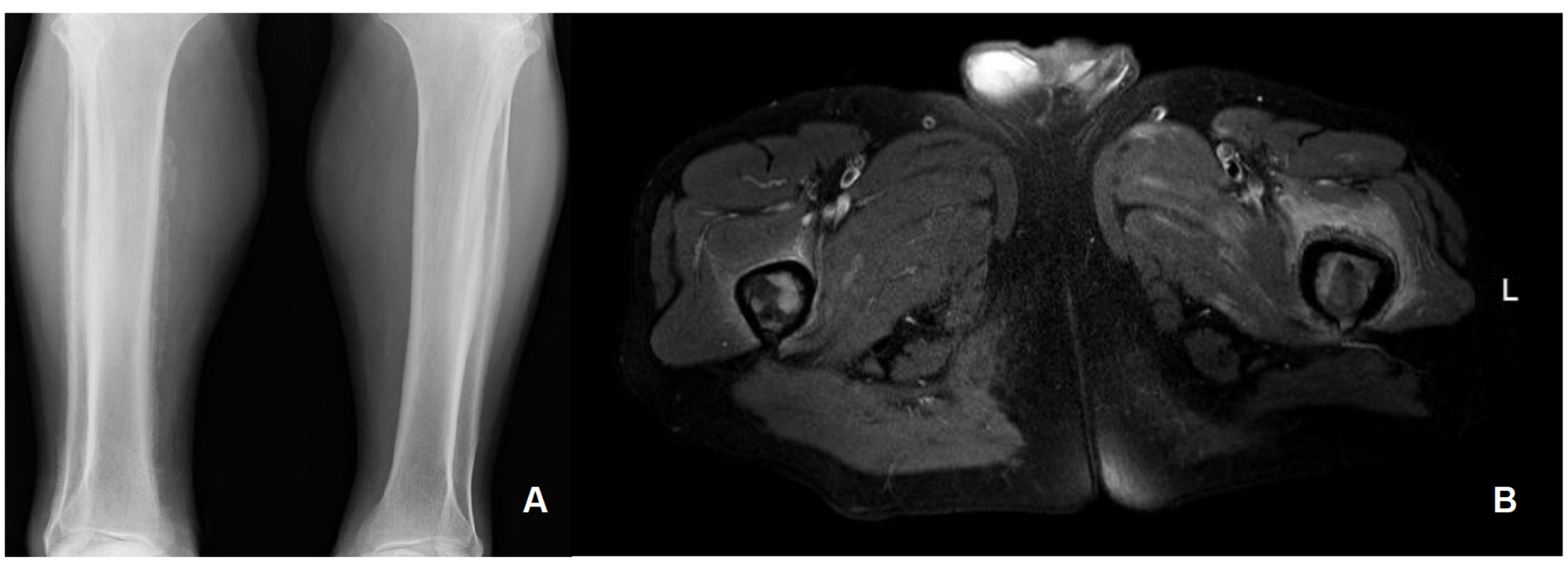
^{99m} Tc Bone scintigraphy: Diffuse increased isotope uptake along the cortical margins of left lower extremety.

CASE

66 years old male patient was admitted to our hospital with pain and swelling of his left lower extremity for 2 months. The pain was intractable leaving the patient bedridden. He denies any other systemic symptoms such as fever and weight loss. He had history of hypertension and aortailiac bypass surgery for aortic aneurysm 5 years ago. On physical examination there were pitting edema on his both legs, predominantly left, no hyperemia or temperature increase was found. There were no pathologic findings during examination of other systems except clubbing of the toes. His bone scan revealed increased osteoblastic activity on left anterior iliac spine and also diffuse expansive heterogen osteoblastic activity was seen on left humerus, tibia, fibula and digital bones. This involvement was decided as Paget's disease although alkalenphosphatase was mildly elevated and he was treated with zoledronic acid 5mg parenterally in another center. On his admission to our hospital, periostal reaction of bilateral femur and tibia was seen on plain radiographs. HOA was decided as preliminary diagnosis. Positron emission tomography (PET) and thorax computerized tomography (CT) were performed in order to identify the etiology of HOA. There were no pathology on thorax CT while PET CT revealed bilateral heterogen periostal reaction of lower extremities confirming HO. Also there was increased FDG activity on abdominal aorta and iliac bifurcation indicating aortitis. Contrast enhanced abdominal CT showed periaortitis caused by a fistula between aortic graft wall and 3. part of the duodenum. The fistula tract was not connected to vascular lumen. Daptomicine 350mg/d was started after obtaining blood cultures. Ertapenem 1gr/d was added due to increased fever during follow-up. Revision graft operation was decided during cardiovascular surgery consultation. He will be operated after 6 weeks of antibiotic treatment is completed.

CONCLUSIONS

Secondary HOA develops as a consequence of various diseases, mainly intrathoracic malignancies. Vascular graft infection, as reported here, is a rare cause of HOA but it might be strong and important sign of infection of vascular prosthesis.



A. Plain X-ray ,
B. MRI of the patient showing periostitis and surrounding soft tissue inflammation and edema.