

A Case of Pathological Rib Fractures: Focal Osteolysis or Osteoporosis?

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ABSTRACT

This paper reports on a unique, previously unreported, successful outcome in the case of a patient with focal osteolytic lesions of the ribs as a first sign of osteoporosis. The lesions were detected by chance after acute cough-induced rib fractures were seen on plain chest radiographs. The diagnosis had to be approached as a diagnosis of exclusion since known causes of the osteolytic process had to be eliminated. The authors describe multiple focal osteolytic lesions with rib fractures appearing in a pattern that could be confused with metastases. Laboratory results were normal. Final diagnosis was based on plain radiography, bone scan and bone densitometry. Pharmacomedical treatments for osteoporosis were applied. The patient was observed between the year 2000 and 2005. Five years later, radiological and bone scintigraphy revealed resolution of the lesion. We conclude that osteoporosis should be included in the differential diagnosis of asymptomatic focal osteolysis of the ribs with rib fractures as a complication of acute cough. The case suggests that focal osteolytic lesions of the ribs may regress over time and become scintigraphically inactive.

Un Caso de Fracturas Patológicas de Costillas: Osteólisis Focal u Osteoporosis Focal?

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RESUMEN

Este trabajo presenta la evolución clínica exitosa, única, y no reportada con anterioridad, del caso de un paciente con lesiones osteolíticas focales en las costillas, como primera señal de osteoporosis. Las lesiones fueron detectadas por casualidad, luego que en unas radiografías simples de tórax, se observaran fracturas de las costillas inducidas por tos aguda. El diagnóstico tuvo que ser abordado como diagnóstico de exclusión, ya que las causas conocidas del proceso osteolítico tenían que ser descartadas. Los autores describen las lesiones osteolíticas focales múltiples con fracturas de costilla presentes en un patrón que podría confundirse con metástasis. Los resultados de laboratorio fueron normales. El diagnóstico final se basó en la radiografía simple, escaneo óseo y densitometría ósea. Se aplicaron tratamientos fármaco-médicos para la osteoporosis. El paciente estuvo sujeto a observación entre el año 2000 y el 2005. Cinco años más tarde, la centellografía ósea y radiológica reveló la resolución de la lesión. Concluimos que la osteoporosis debía incluirse en el diagnóstico diferencial de la osteólisis focal asintomática de las costillas con fracturas de costillas, como complicación de la tos aguda. El caso sugiere que las lesiones osteolíticas focales de las costillas pueden experimentar una regresión con el tiempo y hacerse centellográficamente inactivas.

West Indian Med J 2008; 57 (2): 178

INTRODUCTION

Osteoporosis is characterized by low bone mineral density, microarchitectural deterioration of bony tissue and a

consequent increase in fracture risk. Current knowledge regarding the basic epidemiology of osteoporotic fractures is largely limited to a few fracture sites, notably those of the thoracolumbar sites with vertebral compression and long bones involving the hip and distal forearm (1).

Rib fractures are not a common fracture site, either in osteoporosis or in osteopenia (1). There are some instances in the literature of cough-induced rib fractures which occur primarily in osteoporotic postmenopausal women with chronic cough (2–9).

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When focal bone loss in ribs is diagnosed, known causes of an osteolytic process should be eliminated. Osteolytic rib lesions on plain chest radiograms and hot spots on bone scan in a patient with extraskeletal malignancy is a relatively common finding and is reported to be associated with metastasis (10, 11). A broad variety of diseases should be considered in the differential diagnosis of focal rib osteolysis.

Reported herein is a previously well premenopausal woman, with no history of trauma without osteoporotic risk-factors and with acute cough-induced bilateral rib fractures as the first sign of osteoporosis.

The diagnosis had to be approached as a diagnosis of exclusion. This combination of clinical findings is quite unusual and to the best of the authors' knowledge no similar case has been documented in the literature.

CASE REPORT

This previously well 50-year old premenopausal woman reported severe chest pain after onset of acute cough (2 weeks duration). There was no history of trauma and no acute respiratory infection. Physical examination showed marked bilateral tenderness on the chest wall during respiration. Regional lymphadenopathy and hepatosplenomegaly were not detected. Skeletal survey by plain chest radiography showed, at that time, multiple focal involvement of ribs with bilateral osteolytic lesions and rib fractures (Fig. 1a, 1b).



Fig. 1a: Photograph of a PA chest X-ray. There appears to be a lytic lesion in the posterior aspect of the right 8th rib. Its margins are widely separated. This does not look like a typical fracture.



Fig. 1b: Oblique view of the right hemithorax showing an atypical fracture of the right 8th rib and radiolucent areas in several other ribs.

A technetium-99m diphosphonate bone scan showed increased radioisotope uptake in the areas identified as abnormal by plain radiography (Fig. 2a, 2b).

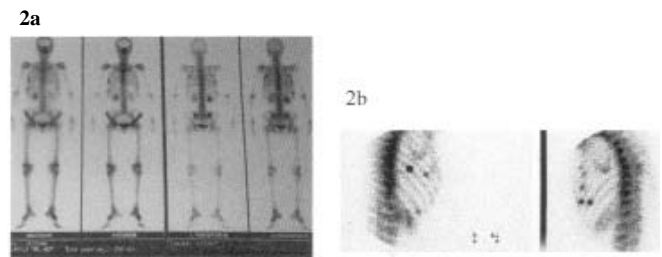


Fig. 2a, 2b: Shows multiple areas of increased uptake in several ribs on scintigraphy. The study dated November 29, 2000.

Laboratory data were at normal range including: erythrocyte sedimentation rate, C-reactive protein, white blood cell count, liver transaminases, serum levels of calcium and phosphate with neutral calcium balance. Also, serum levels of alkaline phosphatase and thyroid and parathyroid hormones were at normal range. Serum protein electrophoresis showed no homogeneous spike in the globulin fraction. Breasts ultrasound and mammography showed no evidence of malignant process. Computed tomography of the chest, abdomen and pelvis revealed no evidence of primary cancer and metastasis. Endoscopy showed no evidence of gastrointestinal lesions. Bone marrow aspiration at the sternum did not reveal pathologic cells. Allergologic tests and tuberculosis tests were negative and the same was the case with bronchoscopy.

To establish the diagnosis, open biopsy of the 8th right rib was performed. The results were negative for tumour cells. Only thin trabecular bone with callus formation was observed.

At that time, on the basis of the historical, clinical, radiological and laboratory evaluation conducted beforehand, the aetiology of the rib lesions was still unknown. The last suspected diagnosis was that of osteoporosis.

A dual-energy X-ray absorptiometry (DEXA) bone densitometry was done and normal mineral bone density was found in the lumbar spine, 1.098 g/cm², with T score: -0.9. An exception was found in the L1, T score: -1.3 and in the L2, T score: -1.7 which revealed osteopaenia. Both hips also showed osteopaenia. The left hip showed T score -1.0 whereas the right hip had T score -1.5. The rate of bone loss was approximately 0.13 g/cm² and 0.18 g/cm² at both hips.

Histological examination of a biopsy specimen obtained from the left iliac crest revealed decrease in bone volume with thin trabecular bone, increase in osteoclastic activity and normal osteoblastic activity. Osteoclasts were not characteristic for Paget's disease of bone.

Alendronate, an orally administered bisphosphonate, was prescribed with supplemental calcium and vitamin D. This regimen was well tolerated and had no gastrointestinal side effects.

After five years of treatment with alendronate, calcium and vitamin D DEXA bone densitometry had improved in all the measured sites: the L1 T score: -0.3, the L2 T score: 0.3 and left hip T score: -0.5. Plain radiography revealed old rib fractures with callus formation and loss of continuity of the 8th right rib on biopsy (Fig. 3a, 3b). Bone scans taken at the

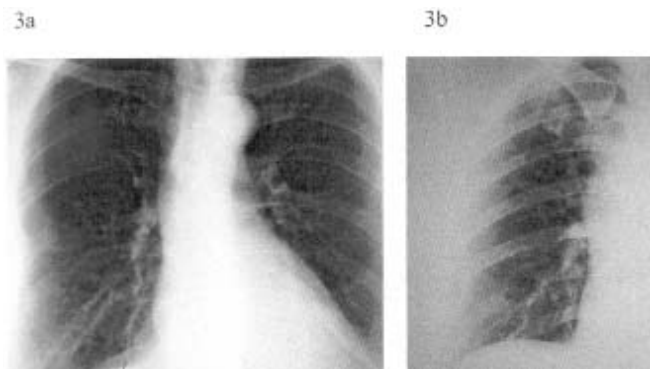


Fig. 3a, 3b: Show photographs of a PA chest X-ray and an oblique view of the right hemithorax, showing the condition following open biopsy of the 8th right rib which was performed 5 years previously and radiological resolution of other rib lesions (decrease in the size of the radiolucent areas).

same time showed decrease in uptake of radioisotope without hot spots (Fig. 4a, 4b).

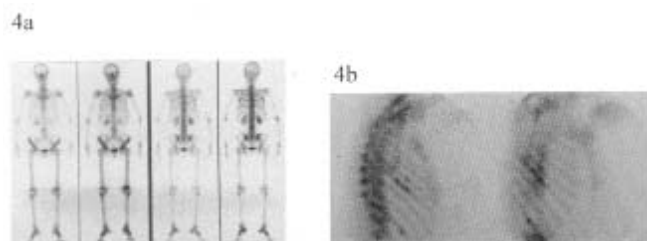


Fig. 4a, 4b: Show no increased uptake in the ribs. This study is dated April 12, 2005.

The patient was observed between the year 2000 and 2005 after which time she was discharged from therapy.

DISCUSSION

Cough induced rib fractures, without any evidence of a malignant process, occur rarely in previously healthy persons and is primarily seen in osteoporotic postmenopausal women (2). They are more commonly associated with chronic cough of known aetiology and are likely to be related to repetitive mechanical stress to the ribs caused by coughing (2). To our knowledge, the study of Hanak *et al* is the only study which presents 54 patients with cough-induced rib fractures. Most of the patients reported by Hanak *et al* who underwent bone densitometry were in the osteopenic and osteoporotic range. However, cough-induced rib fractures can occur in persons with normal bone density (2, 5). This kind of trauma has several similarities to rib fractures induced by rigorous athletic training (12–15). Furthermore,

in elite rowers, rib fractures can occur because the lateral aspect of the rib cage is subject to the greatest bending force during coughing and rowing (12–15). A similar trend is seen in few case reports of cough-induced fractures reported to date (2–8, 12–15). Chest radiography shows a relatively low sensitivity to the detection of cough induced rib fractures and may only reveal up to 60% of the fractures (2, 11, 16). Displaced rib fractures and fractures with callus formation are relatively easy to detect on plain radiographs. In contrast, fractures of ribs that are not displaced or that affect only one aspect of bony cortex (analogous to the greenstick fractures in children) are more difficult to detect with radiography (2, 3). Radionuclide bone scan and CT are more sensitive tools for the detection of stress rib fractures (2, 10, 11, 16, 17). In the reported case, the cough-induced rib fractures occurred in a previously well, premenopausal woman and was associated with acute cough of unknown aetiology. These rib fractures occurred at the lateral aspect of the rib cage because of the greatest bending force during repeated coughing at the sites of bone insufficiency, secondary to unknown cause. Furthermore, in the reported case, chest radiography had a relatively high sensitivity because fractures began in focal osteolytic lesions of the ribs with clearly seen fracture line and bone cortical disruption. Radionuclide bone scans have supported the previously observed specific bone loss bilaterally on the lateral aspect of the rib cage.

Due to the fact that osteolytic rib lesions on plain chest radiograms and hot spots on bone scan are relatively common findings in a patient with extraskelatal malignancy and because they are reported to be associated with metastasis (10), the initial diagnosis was a metastatic process or extraskelatal malignancy. The diagnosis should be approached as a diagnosis of exclusion. Known causes of an osteolytic process should be eliminated. Paget's disease of bone and multiple myeloma should be excluded as underlying causes because urgent treatment of these malignancies is indicated.

Furthermore, multiple randomly distributed foci of increased radionuclide uptake in ribs, mimicking the pattern of osseous metastases, have been reported in several inflammatory, vascular and traumatic conditions. Reported cases include Cushing's syndrome, extreme altitudes, post-traumatic situations, tuberculosis, brucellosis and coccidiomycosis (18–23).

There were no identifiable causes for focal bone loss of the rib. The last diagnosis, still valid, was osteoporosis. Despite the fact that rib fractures are not the common site for osteoporotic fracture, Hanak *et al* in their study reported that reduced bone density is a risk factor and rib fractures could be a complication of chronic cough. However, descriptions of cough-induced rib fractures have been limited to diffuse loss of bone tissue which is radiologically hard to detect. Furthermore, in cases reviewed in the literature, there is no reported case of cough-induced fractures secondary to osteoporosis of the ribs.

In this case, to the date of acute cough, asymptomatic focal osteolysis of the ribs were observed at multiple sites. The lesions were detected by chance after acute cough-induced rib fractures and were seen on plain radiographs of the chest. The unique features of the case are discrepancy among mild densitometry results in the range of osteopenia and radiologically positive focal osteoporosis of the ribs mimicking the pattern of osseous metastases. The patient hardly fitted the diagnostic criteria for osteopenia measured by bone densitometry at the lumbar spine and hips.

Anyway, the treatment with alendronate, calcium and vitamin D was administered and a wait-and-see approach was the only possibility. The substantial improvement in densitometry T scores and decrease in uptake of radioisotope with resolved hot spots in the ribs after five years of treatment suggest that appropriate therapy was chosen.

Having suffered only osteoporotic rib fractures, her disease was relatively mild in comparison with the large spectrum of initial differential diagnosis, especially those associated with the suspicion of bone infiltrating malignancies.

We conclude that osteoporosis should be included in the differential diagnosis of asymptomatic focal osteolysis of the ribs with rib fractures as a complication of acute cough. This case suggests that focal osteolytic lesions of the ribs have a tendency to regress over time and become scintigraphically inactive, as well as that the conservative pharmacological management of osteoporosis was successful.

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