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Case Report

Mammary granuloma revealing multisystemic sarcoidosis

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Abstract

Sarcoidosis is an inflammatory granulomatous disease that can affect any organ system. Axial bone involvement and osteolytic lesion of the spine and pelvic bone was exceptionally reported. That shouldn't be easily diagnosed as osseous sarcoidosis. Also, mammary granuloma was rarely reported as a manifestation of systemic sarcoidosis. Herein, we report a case of a 56-year-old woman who presented with swelling in her right breast. Breast ultrasound showed a nodule of the right breast which histological findings were consistent with granulomatous adenitis. She had also osteolytic bone lesions in the spine without pathological fixation on scintigraphy. Diagnosis of sarcoidosis with uncommon localization: mammary and bone involvement was retained. Such findings in the same patient create a challenge in etiologic diagnosis. Histological confirmation associated to appropriate clinical and radiological presentation are the key of diagnosis.

Abbreviations

CT: Computed Tomography
MRI: Magnetic Resonance Imaging
ACE: Angiotensin Converting Enzyme

1. Introduction

Sarcoidosis is a multi-systemic disease described in the first time by Besnier and al in 1889 [1]. It is characterized by non-caseating granuloma that involves multiple organs in the body affecting the lungs, lymph nodes, liver , spleen, skin, heart, nervous system (neurosarcoidosis), eyes, oto-rhino-laryngeal sphere, locomotor system, heart and mammary tissue [2, 3]. It can be presented with various symptoms depending on organ involvement. The most common signs include coughing, shortness of breath, chest pain, fatigue, weight loss, rheumatologic symptoms, skin lesions. When the disease affects uncommon organs, the diagnosis become difficult like bone and breast involvement. Sarcoidosis with breast mass, axial bone localization was exceptionally reported in the literature [3–5]. 50 % of cases of osseous sarcoidosis was asymptomatic [4]. Less than 1% of patients, sarcoidosis was presented as a mass on the breast with few cases being reported in the literature. Therefore, although rare, a breast mass associated to bone osteolysis should ruling out breast cancer and other differential diagnosis in order to have better prognosis [6, 7]. Herein, we report an exceptional case of multi systemic sarcoidosis affecting the breast tissue, axial bone and lungs.

2. Case report

58-year-old women was admitted in internal medicine department for granulomatous lymphadenitis. In her medical history, we found a chronic severe anemia treated by transfusion, vitamin B9 and iron supplementation with good progress.

Her recent history dates back to May 2024 when she was hospitalized in gastroenterology department for recurrence of anemia. It was a moderate anemia at 10g/dL considered as an iron deficiency anemia. In anamnesis, she didn't report geophagy. She did not describe any externalized bleeding. In order to conduct an appropriate etiological investigation, an endoscopic exploration (esophago-gastroduodenal fibroscopy and colonoscopy) was performed concluding to normal appearance of digestif tract. Thoraco abdominal pelvic computed tomography (CT) showed nodular lesion in the right breast associated to nodular and micro nodular lesions in pulmonary parenchyma and even more multiple osteolytic lesions of the spine. Ultra sound breast showed the nodule in the union of external quadrant of the right breast measuring 13*10 mm; associated with a retro-areolar area. The biopsy of the breast lesion guided by ultrasound was performed found lymphadenitis with non-necrotizing granuloma without histologic signs of malignancy.

Then, she was referred to our department of internal medicine for further investigation.

In clinical assessment, she was presented with preserved general status. She weighed 86 kg. His blood pressure was 120/60 mmHg. Heart and lung auscultation is normal. Physical examination didn't show pathologic findings. Biological analysis found microcytic non-regenerative anemia hemoglobin: 8g/dL with lymphopenia (lymphocytes 1300 /mm3). Kidney function and liver tests were within normal range. The phosphor-calcium assessment found elevated leved urinary calcium with normal blood calcium test (Ca^{2+}: 2,3 mmol/L, calciuria day1:7,5mmol/day and calciuria day2:8mmol/day, Albumin 37, 9g/L). Rereading of computed tomography was performed. Thoracic sections especially parenchymal window showed multiple bilateral nodules and micronodules with peri-lymphatic distribution of which the largest measured the largest measured 6mm localized in the middle lobe Figure 1.

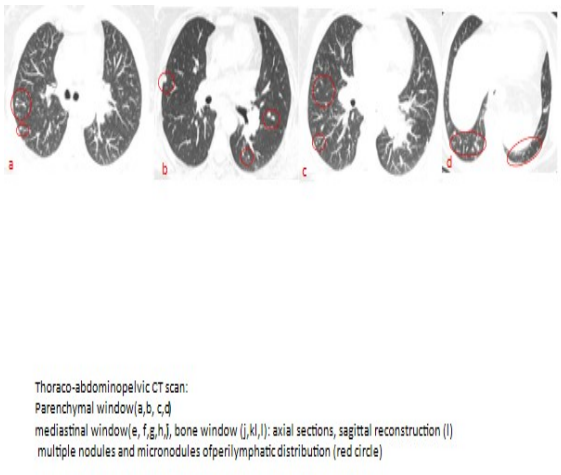

Figure 1: Chest computed tomography (Parenchymal window)

The nodular lesion of the breast figured in Figure 2. In mediastinal window, there was multiple non compressive bilateral mediastinal and inter bronchial lymph nodes of which the largest measured 30*22 mm Figure 2.

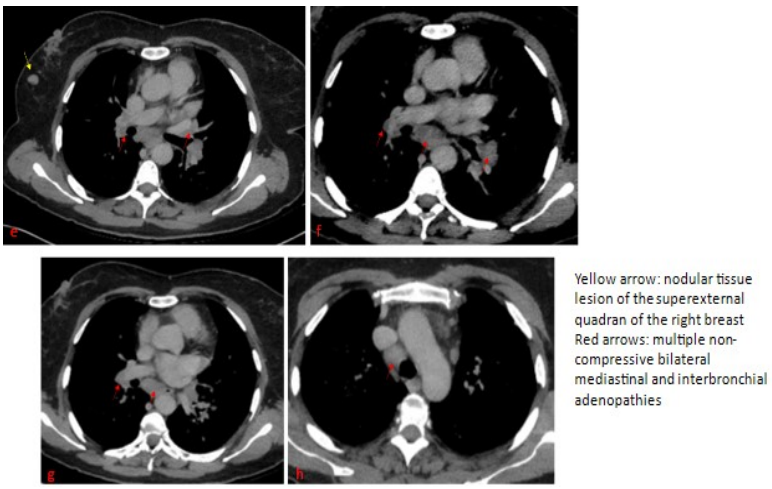

Figure 2: Chest computed tomography (mediastinal window) revealing the nodular tissue lesion of the breast and bilateral swollen lymph nodes

In bone window, multiple well limited osteolytic lesions without peripheral osteosclerosis was objectified concluding as Lodwick type IB with bone cortical fracture in the right iliac wing, distributed over the sternum, ribs, cervico dorso lumbar spine and pelvic bones Figure 3.

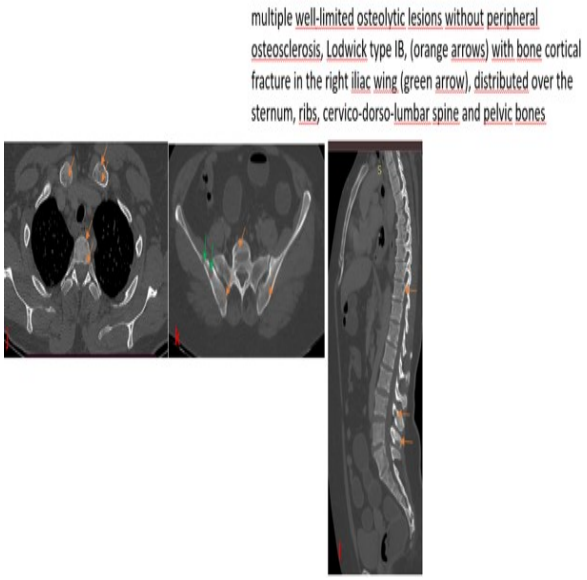

Figure 3: Osteolytic lesions in the axial skeleton

Abdominal sections found heterogenous appearance of the liver and the spleen Figure 4.

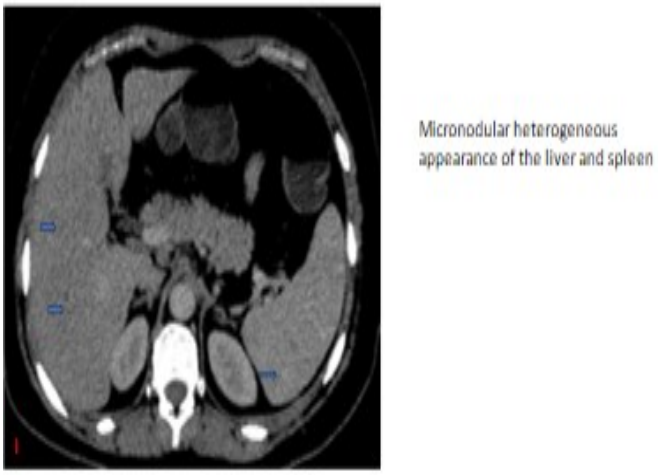

Figure 4: osteolytic lesions in the axial skeleton

Blood tests revealed elevated ACE levels of 100 U/L (20–70 U/L). Accessory salivary gland biopsy didn't found granuloma. The patient underwent bronchial fibroscopy which showed enlarged bronchial spurs. Bronchoalveolar lavage revealed lymphocytosis and bronchial biopsy didn't found malignant cells.

A staging technetium 99m-methyl diphosphonate (99mTc MDP) whole-body bone scan was practiced, didn't show pathologic fixation.

At the end of clinical examination and paraclinical explorations, exclusion of alternative causes was established. The diagnosis of multi systemic sarcoidosis was based on clinical, radiological, biological, and histological criteria. At this time, the patient had no respiratory symptoms and her calcium level was within the normal range. A regular checking was proposed and our patient was put under clinical and radiological surveillance.

3. Discussion

Sarcoidosis is a granulomatous pathology with multi-systemic involvement. It was pathologically manifested by the formation of non-caseating epithelioid granulomas. Rheumatologic manifestations in sarcoidosis varies from arthralgia to bone involvement [8–10]. It occurs in 4% to 38% of patients. They may manifest clinically as inflammatory arthritis, periarticular tissue swelling, tenosynovitis, dactylitis, bone involvement, or myopathy. Oligo and polyarthritis involving the ankles may be considered suggestive of rheumatic sarcoidosis.

Osseous sarcoidosis is rare. In a cohort of 100 patients, osseous sarcoidosis was more common in females during the fifth decade of life [8]. Osseous lesions , like the case of our patient, are found in up to 13% of patients with sarcoidosis [4, 11, 12]. Most patients with vertebral involvement have a known diagnosis of intrathoracic sarcoidosis [6]. While the peripheral skeleton was a frequent location, data regarding axial involvement is scarce. It was frequently asymptomatic and was incidentally detected in 50% [6]. Dactylitis and osseous pain were the first main clinical symptoms.

In X-ray radiography, the pattern of axial involvement varies from purely lytic to mixed lytic-sclerotic and purely sclerotic lesions [11]. Our patient had lytic lesions Lodwick IB atypical for osseous sarcoidosis. Magnetic Resonance Imaging (MRI) was important in cases of axial sarcoidosis to helps in order to exclude differential diagnoses and delineate the extent of bone involvement. Although sarcoidosis of the bone is rare and is encountered much less frequently than skeletal metastases, it is important to consider it in the differential diagnosis in a patient with breast nodule given the similarities in imaging findings with divergent clinical courses [11]. Sarcoidosis should always be considered as an elimination diagnosis because of it low incidence. Less commonly, in around 1% of cases, sarcoidosis presents with a breast mass. In the literature, only 35 cases were identified between the years 1921 and 1997 that histologically proved the existence of sarcoidosis in the breast. Only some cases reports were described in the literature [4, 7]. Breast masses, even in patients without clinical evidence of systemic sarcoidosis are rarely linked to systemic sarcoidosis. When the diagnosis of mammary granuloma is suspected by fine needle aspiration cytology, exceptional procedures should be also considered to examine for the possibility of a coexisting carcinoma, which may have been missed as a result of a sampling error [7, 12–14]. In such cases, excisional biopsy or resection is strongly recommended [6]; In our patient, non-necrotizing granulomas were found in lymph node of the breast which was unhabitual finding.

Article Information

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Disclaimer (Artificial Intelligence): The author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc.), and text-to-image generators have been used during writing or editing of manuscripts.

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