

ACUTE PRIMARY TUBERCULOUS MASTITIS: A CASE REPORT

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ABSTRACT

This is a report of a 28-year-old female patient neither pregnant, nor lactating and was previously healthy, not known to be immunocompromized who presented with a two month history of a painful lump in her right breast associated with low-grade fever. A thorough physical examination, laboratory and radiological investigations were done including breast mammogram, breast ultrasound and excisional biopsy from breast mass for histopathological study; that showed caseating epithelioid granulomas with giant cell and the Ziehl-Neelsen stain was negative.

Key words: Tuberculosis, Primary, Mastitis

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Introduction

Acute secondary tuberculous mastitis is a rare disease mainly affecting young women, most often during pregnancy and/or lactation. This entity was first described by Sir Astley Paston Cooper in 1829 as "scrofulous swelling in the bosoms of young women most of whom suffered from tuberculous cervical adenitis" and further elaborated by Cohen in 1977.⁽¹⁻³⁾ Approximately 550 cases have been described in the English literature, as secondary tuberculous mastitis, but primary tuberculous mastitis is extremely rare and till now a few cases have been reported internationally using a Medline search since 1960. Many pathologists and clinicians are still unaware of this disease entity. Clinically, the condition may closely mimic carcinoma, and histopathologic appearance, if not recognized as granulomatous disease, may be misinterpreted as a carcinoma, often with disastrous consequences.⁽⁴⁾

Case Report

A 28-year-old woman, previously healthy, non-smoker, neither pregnant, nor lactating, having oligomenorrhea diagnosed as polycystic ovary syndrome and treated with Metformin 850mg twice daily was referred to the Internal Medicine Clinic at Prince Rashed Bin Al-Hassan Hospital (PRHH), Jordan on 22nd of September 2005. She presented with a painful mass lesion in the right breast, which has been present since 25th July 2005, coinciding two weeks after the return of her healthy, asymptomatic and thoroughly investigated husband from Eritrea. She complained of severe itching in her right breast around the areola, hotness and spikes of fever. On examination, she looked healthy, well built, well nourished but anxious about her condition. Blood Pressure was 110/70mmHg, pulse rate 110 beats per minute, oral temperature was 37.9°C. Systemic examination revealed no

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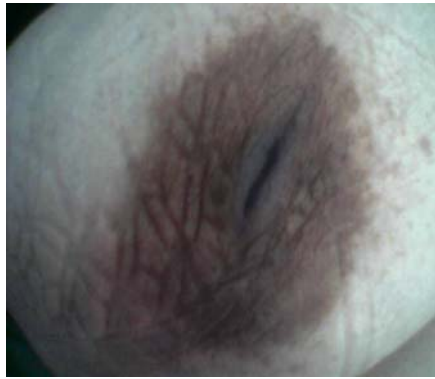


Fig. 1. The nipple was retracted, showing sinus formation, the skin was hot, red and edematous with a peau d'orange appearance

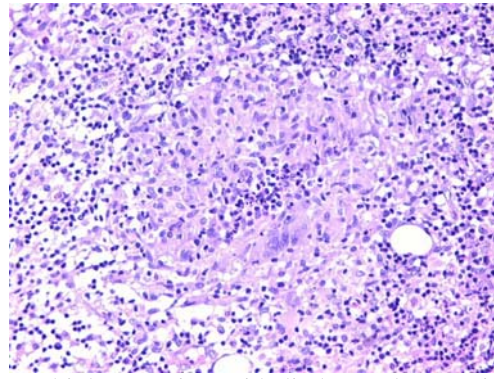


Fig. 2. Multiple caseating epithelioid granulomas with Langhan's giant cells surrounded by severe acute and chronic inflammation

significant abnormalities. No cervical or axillary lymphadenopathy, chest and heart examination was normal. On breast examination the nipple was retracted, showing sinus formation, the skin was hot, red and edematous with a peau d'orange appearance (Fig. 1). A huge, firm and tender 10x10cm nodule was palpated in the upper outer quadrant of the right breast. Abdominal examination was normal; no ascitis or hepatosplenomegaly were found. Examination of upper and lower extremities was normal, skin examination was normal.

Complete Blood Count, Prothrombin Time, activated Partial Thromboplastin Time, Liver Function Tests (LFT), Kidney Function Tests (KFT), Electrocardiogram (ECG), Chest-X-Ray (CXR), Urinalysis, Urine culture and abdominal ultrasound were normal.

Tuberculin test, Human Immunodeficiency Viral (HIV) and the Venereal Disease Research Laboratory Slide (VDRL) tests were negative; also the Brucella test was negative. IgG and IgM antibodies against both Toxoplasma and Cytomegalovirus serology were negative as a remote possibility of this condition. Erythrocyte Sedimentation Rate (ESR) was 56mm in the first hour. Mammogram showed focal asymmetrical dense lesion in the upper outer quadrant of the right breast. Breast ultrasound showed an echogenic right upper outer quadrant dense lesion with no collection. The provisional diagnosis of breast abscess or malignancy was suggested and the patient was admitted to hospital for fine needle aspiration (FNA) and excisional biopsy. FNA revealed sheets of polymorphs and histiocytes in a hemorrhagic background showing collection of epithelioid cells

and multinucleated giant cells; the appearances were those of an inflammatory granulomatous process. While the histopathological study revealed multiple caseating epithelioid granulomas with Langhan's giant cells surrounded by severe acute and chronic inflammation, the Ziehl-Neelsen (ZN) stain for acid fast bacilli (AFB) was negative for the time being, however the features were those of tuberculous mastitis (Fig. 2).

Pathological brownish fluid was extracted on three occasions from the mass lesion and tested for tuberculosis with the Polymerase Chain Reaction (PCR); the primers used were AMS19 by amplification of IS6110 region, the amplification product is a 317bp which codifies the IS6110 region and it is detected by electrophoresis on (pre-cast) clearance gel containing ethidium bromide (a production of CLONIT S.r.l, (Milan, Italy) and the last test was positive for tuberculosis.

The patient was started on anti-tuberculosis therapy for 6 to 9 months. Also we investigated her husband; a 30-year-old soldier, smoker, healthy and asymptomatic, his physical examination was unremarkable, chest X-Ray, CBC, KFT, LFT, urinalysis and urine culture were normal. Tuberculin test was positive, suggesting the "possibility" that her husband could be the source of infection, but unfortunately we lost his follow-up.

Discussion

Tuberculosis of the breast is extremely rare and occurs in young, multiparous lactating women. Its incidence in western countries is less than 0.1% of breast lesions examined histologically and 3% to

4.4% of all breast diseases treated in the developing world where TB is endemic.⁽⁵⁾ On a clinical and pathological bases five different patterns of tuberculous mastitis are described: acute miliary tuberculosis mastitis, nodular tuberculosis mastitis, disseminated tuberculosis mastitis, sclerosing tuberculosis mastitis and tuberculosis mastitis obliterans.⁽⁶⁾ Clinically, the above groups of lesions can be mistaken for neoplasms. Nodular tuberculosis can be mistaken for fibroadenoma or carcinoma while disseminated tuberculosis can be mistaken for inflammatory carcinoma, and sclerosing tuberculosis can produce extensive fibrosis and nipple retraction, being easily mistaken for scirrhous carcinoma.^(4,7)

At present, mammographic studies in addition to fine needle aspiration studies are not helpful for the diagnosis of tuberculous mastitis. An excisional biopsy is the most reliable for the definite diagnosis of tuberculous mastitis.⁽⁸⁾ The diagnosis, however, is very difficult, especially in a number of pathologic processes such as comedo-mastitis, which with its chronic inflammatory cells, is similar to tuberculosis. In these situations, the demonstration of acid-fast bacilli is mandatory for definite diagnosis. Overall, male breast tuberculosis is very rare and only few male breast tuberculosis cases have been reported.⁽⁶⁾ In this case, many differential diagnoses were considered: breast abscess, acute mastitis, sarcoidosis, fat necrosis, papilloma, fibroadenoma and carcinoma. The excisional biopsy material revealed numerous caseating epithelioid granulomas with Langhan's giant cells surrounded by severe acute and chronic inflammation of the breast; those of tuberculous mastitis.⁽⁵⁾

There are three recognized modes of spread of the tubercle bacilli to the breast: direct, lymphatic and haematogenous. Rarely, infected sputum can reach the underlying breast through superficial abrasions of the skin of the breast. In all cases bacilli infect the ducts and spare the lobules. Dilated ducts of the breast in pregnant and lactating women appear to be especially susceptible to infection. Retrograde spread of infection from primary foci of disease in the lymph node (axillary, mediastinum, parasternal or cervical) to the breast is also well recognized.^(3,6)

The management of this case was surgical and medical with anti-tuberculous therapy. A wedge resection was performed with a daily dressing but

larger lesions should be treated with simple mastectomy. Extra pulmonary TB is treated like pulmonary TB. A 6-month regimen is thought to be adequate; in the first two months of therapy we start quadruple anti-tuberculous medication: Isoniazide (INH) 300mg/d, Rifampicin (RMP) 600mg/d, Pyrazinamide (PZA) 500mg twice/d and Streptomycin (STM) intramuscular injection 1g/d for the first two weeks. After two months INH and RMP are given up to six months. Treatment of TB mastitis is best achieved by conservative surgery supported with anti-tuberculosis drugs.

Upon follow-up two months later signs of recovery were noted. Her physical examination was unremarkable, both breasts ultrasound was normal, FNA follow-up was also normal. Hence, the combination of drug therapy and limited excision of diseased breast tissue is a method of choice.⁽⁸⁾

Conclusion

TB can affect virtually any organ system in the body and can be devastating if left untreated. Uncommon sites and ability to mimic other diseases clinically and radiographically leads to diagnostic and therapeutic delay. A high level of suspicion is required especially in high-risk populations and endemic areas.

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