Granulomatous mastitis – a rare cause of erythema nodosum

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Summary: Granulomatous mastitis is a recently described, rare condition of unknown aetiology. It occurs in young, parous women as a tender extra-areolar breast lump and therefore needs to be differentiated from carcinoma. Histologically, a discrete granulomatous lobulitis is seen and because of morphological similarities to granulomatous thyroiditis it has been suggested it may be immunologically mediated. We present a patient in whom granulomatous mastitis presented with peri-arthritis and erythema nodosum, an association not previously described.

Introduction

Granulomatous mastitis is a recently described disease with distinctive histological features. The diagnosis depends on the exclusion of other granulomatous diseases including tuberculosis, sarcoidosis or a foreign body reaction.^{1, 2, 3}

The condition presents as a tender extra-areolar breast lump, usually within 2 years post-partum. The lump is often extremely hard and may be mistaken for carcinoma. Systemic features, such as fever, sweats and weight loss, are common but erythema nodosum and peri-arthritis have not been described. Histologically there is a discrete non-caseating granulomatous lobulitis, unlike that seen in tuberculosis or sarcoidosis. The granulomata often coalesce and form microabscesses.

The aetiology is unknown. An allergic basis has been postulated⁴ and the presence of microabscess formation suggests an infective cause although no agent has been isolated.

We present a patient who developed granulomatous mastitis which was complicated by erythema nodosum and peri-arthritis.

Case report

A 24 year old woman gave a 6 week history of a painful lump in the left breast, a 2 week history of tender, red, lumps on both shins and arthralgia affecting hands, knees and ankles. The breast lump developed 4 months after the birth of her second child. Neither child was breast-fed. There was no nipple discharge.

She had not taken the contraceptive pill, or any other medication.

On examination she was pyrexial (38°C). The left breast contained a hard, tender mass 12 × 9 cm in the lower half with indrawing of the nipple. She had florid erythema nodosum affecting both legs and peri-arthritis involving both ankles. There was no lymphadenopathy. An open breast biopsy was performed; the lump was not completely excised.

Macroscopic examination revealed multiple small abscesses. Histology showed areas of inflammation, predominantly related to breast lobules but also involving and partially destroying small ducts and ductules. Inflammatory areas included several granulomata and foci of microabscess formation. The changes were characteristic of idiopathic granulomatous mastitis.

No organisms were seen on Gram, periodic acid-Schiff or Grocott stained sections. However Ziehl-Neelson staining revealed a total of four acid-fast bacilli (AFB).

She had a normal haemoglobin and white cell count, a raised erythrocyte sedimentation rate (104 mm/h) and C-reactive protein (374 mg/ml). Biochemical profile was normal (including calcium and liver function tests). Routine cultures of sputum, urine, and breast tissue were negative. Cultures of breast tissue for mycobacterium by guinea-pig inoculation were negative. Anti-streptolysin O titre was <200 Todd units. Chest X-ray was normal. There was no evidence of immune dysfunction. Serum immunoglobulins were normal, tests for autoantibodies, circulating immune complexes and activated lymphocytes were negative. Tuberculin skin testing was negative at 100 units per ml on admission and when repeated 6 weeks later. A Kveim biopsy was also negative.

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Treatment with indomethacin produced a rapid resolution of the fever, erythema nodosum and arthritis. Because of the presence of AFB in the biopsy specimen she was treated with rifampicin and isoniazid for 6 weeks until the negative culture results were obtained. This was not associated with any significant reduction in the size of the mass. A sterile discharge from the wound persisted for 6 weeks. Thereafter the lump gradually became smaller with almost complete resolution at 5 months.

Discussion

Our patient had the typical clinical profile of idiopathic granulomatous mastitis as described by Kessler & Wooloch, namely the development of a very hard, tender, breast lump post-partum.¹ The timing of the discharge and the slow resolution are characteristic of granulomatous mastitis. The histological features were also typical.

The finding of four AFB was unexpected and difficult to explain. Tuberculosis was excluded by the lack of response to anti-tuberculous therapy, the negative Mantoux (particularly in the presence of erythema nodosum), atypical histology, a normal chest X-ray and, in particular, negative culture results following guinea-pig inoculation with breast tissue. Atypical mycobacterium infection is a possible explanation but the negative culture results make it unlikely. Sarcoidosis may cause granulomatous mas-

titis but the negative Kveim test and lack of other typical features, including histology, fail to support this diagnosis.

Erythema nodosum has not previously been described in conjunction with granulomatous mastitis. It provides supportive evidence for an altered immune status previously suggested as the cause of granulomatous mastitis.^{1,4}

Little is known about the optimal treatment of this condition. Wide excision has been advocated,⁴ although the high incidence of post-operative infections is well documented.³ The use of corticosteroids has been proposed.⁵ However, our patient made a good recovery with only symptomatic treatment. This suggests the disease may be self-limiting and that potentially hazardous treatments should be reserved for the more protracted case.

Granulomatous mastitis should be considered in the differential diagnosis of breast lumps occurring post-partum. The development of erythema nodosum demands the rigorous exclusion of tuberculosis and sarcoidosis. However, from our case it would appear that erythema nodosum may occur in association with idiopathic granulomatous mastitis.

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