Letters to the Editor

Mastitis refractory to cyclophosphamide in systemic lupus erythematosus

Sirs,

Mastitis is not infrequent in women, particularly in the child-bearing age. Most cases are caused by infections; mastitis caused by SLE is a rare condition (1).

In January 2000, a 16-year-old woman was referred for evaluation at the rheumatology unit of the State University of Campinas. She complained of polyarthralgia, weight loss, photosensitivity and oral ulcers. Laboratorial tests showed positive antinuclear antibodies (Hep-2) at 1:2560, with homogeneous and peripheral patterns; positive anti-dsDNAat 1:160 and anti-SSA/Ro. Diagnosis of SLE was made and the patient was put on prednisone 5 mg/day and chloroquine diphosphate 250 mg/day. In March 2002 an indurate mass was felt in her right breast. An ultrasound of her right breast showed multiple nodular areas, ranging between 15 and 40 mm in diameter, with inner echoes, and ductal ectasia. Aspiration yielded a thick, white-yellowish fluid with absent of malignant cells on cytology and negative cultures. It was considered an abscess with partial resolution, and oral ciprofloxacin for 14 days was prescribed without improvement. Surgical drainage was indicated, and bromocriptine and cephazolin was prescribed. After drainage a brief improvement was observed but one month later the area of induration increased again, reaching 8 cm in diameter. An ultrasound showed diffuse parenchyma texture, subcutaneous thickening and collection of thick fluid in the lower quadrants. Given the lack of response to antibiotics, various negative cultures and the relapsing nature of the lesion, a surgical debridement was done. The biopsy showed non-specific chronic mastitis and steatonecrosis; cultures for bacteria, mycobacterium and fungi were negative. Fifteen days after surgery, the affected area was still increasing in size with a great amount of purulent discharge. A new ultrasound showed abscesses involving the majority of the right breast, and a new surgical debridement was done. Since no other etiology factor was identified, we considered the hypothesis of lupic mastitis and prednisone 1 mg/Kg/day, cyclophosphamide 100 mg/day and chloroquine were prescribed. After 4 months no improvement was noted. Considering the extent of the lesion and the lack of treatment efficacy, a partial mastectomy was realized. Histopathological analysis of the surgical specimen evidenced chronic mastitis, with lymphocyte and plasmocyte infiltrate rich in xanthomatous hystiocytes. Steatonecrosis, reactional ductal hyperplasia and a skin fistula were observed.

Lupic mastitis is rare manifestation of lupus erythematosus. In the literature there were only 7 (2, 3-6) reported cases. It is considered a form of lupus panniculitis, also called lupus erythematosus profundus, characterized by inflammation of the subcutaneous fat (7). There are 4 reported cases of mastitis occurring in patients with discoid lupus erythematosus (3, 4, 6) and 3 other cases in SLE (2, 3, 5).

Two to 3% of all patients with SLE present lupus panniculitis, the most common affected places are arms, buttocks, head, neck and thighs. The characteristics of breast involvement are similar to lupus panniculitis elsewhere. It is initially manifested as persistent, well-defined, subcutaneous, solitary or multiple nodules. The lesions may progress to chronic ulcers with chronic discharge or resolve with atrophic scars. Biopsy usually reveals hyaline fat necrosis, lymphocytic infiltration, microcalcifications and lymphocytic vasculitis.

The course of lupic mastitis tends to be chronic and relapsing. The reported treatment regimens vary and include corticosteroids (2), chloroquine (3, 4), cyclophosphamide (3) and mastectomy (5). Our patient received high dose prednisone, chloroquine and cyclophosphamide because of the progressive increase in size of the lesion. Unfortunately, this treatment did not improve lesion size and mastectomy was performed. Surgical procedures are reported to exacerbate the local inflammatory process (4, 5), but the drainages were necessary to alleviate the pain and to rule out infection and malignant etiologies.

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Table I. SLE mastitis. Review of the literature.

Author	Age	Race	Type of LE	Location	SLE features	Treatment
De Bandt M *et al.* (2)	21	Black	Systemic	Both breasts	Serositis, hemolytic anemia, antiphopholipid antibodies, stroke	Oral prednisone, pulses methylprednisolone
Cernea *et al.* (6)	33	White	Discoid	Right arm, left breast	Alopecia	Chloroquine, prednisone
Holland *et al.* (3)	26	Black	Systemic	Lower and upper extremities, right breast	Arthritis, malar rash, nephritis	Prednisone, cyclophosphamide
Holland *et al.* (3)	49	Black	Discoid	Lower and upper extremities, right breast	Discoid lesions	Hydroxychloroquine
Harris *et al.* (4)	36	Black	Discoid	Left breast	Discoid lesions	Chloroquine
Harris *et al.* (4)	70	White	Discoid	Both breasts	Discoid lesions	Hydroxychloroquine
Georgian-Smith *et al.* (5)	44	NA	Systemic	Left breast	Nephritis	Prednisone, mastectomy
Castro *et al.*	21	Afro-brazilian	Systemic	Right breast	Oral ulcers, nephritis,	Prednisone, chloroquine, cyclophosphamide, partial mastectomy

NA: Not available