

Gestational gigantomastia in systemic lupus erythematosus

Ana Luisa S. Pedreira, Mittermayer B. Santiago

Hospital Santa Izabel, Salvador, Bahia, Brazil
Escola Bahiana de Medicina e Saúde Pública, Salvador, Bahia, Brazil
Serviços Especializados em Reumatologia da Bahia, Salvador, Bahia, Brazil

A 33-year-old woman was diagnosed with systemic lupus erythematosus (SLE) 9 years earlier, based on clinical findings of photosensitive malar rash, alopecia, arthritis, and the presence of autoantibodies, such as antinuclear antibodies with a titer of 1:160 and a speckled pattern, as well as anti-double stranded DNA, anti-ribonucleoprotein, and anti-Sjögren syndrome A antibodies.

The patient was regularly using hydroxychloroquine (400 mg/d), with no signs of disease activity or other comorbidities. She became pregnant, and after 10 weeks, she noticed a rapid diffuse, bilateral, and symmetrical enlargement of her breasts, associated with local pain and hyperemia. Her previous pregnancy 10 years earlier was uneventful. With a presumptive diagnosis of

infectious mastitis, she was administered cephalexin (2 g/d) for 10 days, without improvement.

On physical examination, at 15 weeks of pregnancy, she had large breasts with a *peau d'orange* aspect (FIGURE 1), without nodules or nipple discharge, and a chest diameter of 133 cm. The rest of the physical examination was unremarkable. The levels of estradiol, luteinizing hormone, progesterone, prolactin, testosterone, thyrotropin, and T₄ were within the reference ranges. Breast ultrasound showed diffuse thickening of the skin and thickening of the subcutaneous tissue without evidence of organized collection. The patient underwent a deep biopsy of the breasts. Histopathological findings were pseudoangiomatous stromal hyperplasia, apocrine metaplasia, and



FIGURE 1 Bilateral gestational gigantomastia in a patient with systemic lupus erythematosus

Correspondence to:
Mittermayer B. Santiago, MD,
Serviços Especializados em
Reumatologia da Bahia,
Rua Conde Filho 117, Graça,
Salvador, Bahia, Brazil,
CEP 40.150-150,
phone: 71-30229886,
email: mbsantiago2014@gmail.com
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skin with mild lymphocytic vasculitis. Immunohistochemistry was negative for neoplasia. Based on these findings, we diagnosed inflammatory gestational gigantomastia in the context of SLE, a condition characterized by an increase of more than 1.5 kg per breast. The pathogenesis of gigantomastia is poorly understood. However, an increased prevalence of this condition in puberty or during pregnancy highlights the potential role of sex steroids.

Dancey et al¹ performed an extensive review of the literature and proposed a classification for gigantomastia. A total of 108 patients were identified, and the etiology was defined as follows: juvenile in 57 cases, pregnancy-induced in 41 cases, idiopathic in 13 cases, and drug-induced in 4 cases. Ten patients were noted to have concurrent immune diseases.¹ Recently, 50 patients with gestational gigantomastia were analyzed in a systematic review, the majority of whom had bilateral involvement and had their onset in the first or early second trimester of pregnancy.² Gestational gigantomastia was reported in 2 patients with SLE and in 1 patient with undifferentiated connective tissue disease.^{3,4} Another case occurred in a patient with SLE who was neither pregnant nor peripubertal.⁵

The involvement of autoimmunity in gestational gigantomastia is controversial, and no specific antibody against breast tissue antigens has been identified. This disorder is responsible for negative physical and psychosocial effects, and shows an unsatisfactory response to drug therapies. Our patient developed a progressive and exponential increase in the size of her breasts in the course of her pregnancy. She also presented with dyspnea and severe breast pain, as well as with areas of skin necrosis secondary to an inadequate vascular supply. A radical bilateral mastectomy was performed at the 26th week of gestation and the total weight of each breast was 7.5 kg. The histopathology of the breast showed a similar pattern to the previous biopsy. No further clinical complications were reported either for the patient or the neonate after delivery.

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