Turkish Journal of Medical Sciences

Volume 47 | Number 5 Article 39

1-1-2017

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TÜBİTAK

Turkish Journal of Medical Sciences

http://journals.tubitak.gov.tr/medical/

Research Article

Turk J Med Sci
(2017) 47: 1590-1592
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doi:10.3906/sag-1611-100

Coexistence of idiopathic granulomatous mastitis and erythema nodosum: successful treatment with corticosteroids

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Received: 23.11.2016 • **Accepted/Published Online:** 12.02.2017 • **Final Version:** 13.11.2017

Background/aim: Idiopathic granulomatous mastitis (IGM) is a rare, chronic inflammatory disease of the breast. Erythema nodosum (EN) is a rare extramammary manifestation of IGM. The purpose of this study is to determine the clinical and demographic characteristics of 11 IGM and EN patients and to evaluate the efficacy of methylprednisolone treatment.

Materials and methods: In our series, ten patients had EN bilaterally, whereas one patient had a lesion of the right pretibial area. The mean age of the patients was 35.5 years (range: 29–45 years). IGM and EN were diagnosed by the necessary serological, microbiological, radiological, and histopathological examination. After diagnosis, methylprednisolone was started in the first week at 0.8 mg/kg daily for treatment. The weekly dose was tapered to 0.1 mg/kg daily over 8 weeks.

Results: We started with the treatment of methylprednisolone, and in all our cases the initial response was excellent. In 2 weeks the IGM symptoms had markedly declined, while signs of EN disappeared completely. Patients were followed for an average of 60 months after treatment. None of the 11 patients had recurrence.

Conclusion: We herein report a rare series considering IGM cases complicated by EN. Few such cases have been reported in the literature. We advocate for an initial trial of methylprednisolone treatment, which proved to be very successful in our patients.

Key words: Granulomatous mastitis, idiopathic granulomatous mastitis, erythema nodosum, breast, corticosteroids

1. Introduction

Granulomatous lobular mastitis (GLM) is a rare inflammatory breast disease (1). It usually presents with an irregular circumscribed breast mass, breast pain, nipple inversion, cutaneous ulceration, induration, erythema, and cutaneous fistula formation. Its radiological and clinical symptoms are similar to breast carcinoma, so unnecessary surgical procedures are likely to be applied. There are two forms of GLM: idiopathic granulomatous mastitis (IGM) and specific granulomatous mastitis. Parasitic infections with specific effects, tuberculosis, Wegener granulomatosis, sarcoidosis, histoplasmosis, foreign bodies, and fungi should be excluded in potential cases of IGM (2). Erythema nodosum (EN) is the most common type of panniculitis and is characterized by reddish, painful nodules in the subcutaneous area. It is typically located in pretibial regions bilaterally (3). EN is a rare systemic manifestation of IGM. The relationship between IGM and EN was first described by Adams in 1987 (4). The purpose of this study is to determine the clinical and demographic characteristics of 11 IGM and EN patients and to evaluate the efficacy of methylprednisolone over a long follow-up period.

2. Materials and methods

Eleven patients who were admitted to the General Surgery Outpatient Clinic of the Gazi University Faculty of Medicine between 2006 and 2015 with complaints of breast mass and cutaneous fistula formation, who were diagnosed with IGM (Figure 1A) with concurrent varying numbers of pretibial subcutaneous nodular lesions consistent with EN (Figure 1B), were selected. Abscess and tissue cultures were obtained from the patients for microbiological examination. Antibiotic therapy was planned according to these results. Acid-resident bacteria, Löwenstein–Jensen culture, and tissue polymerase

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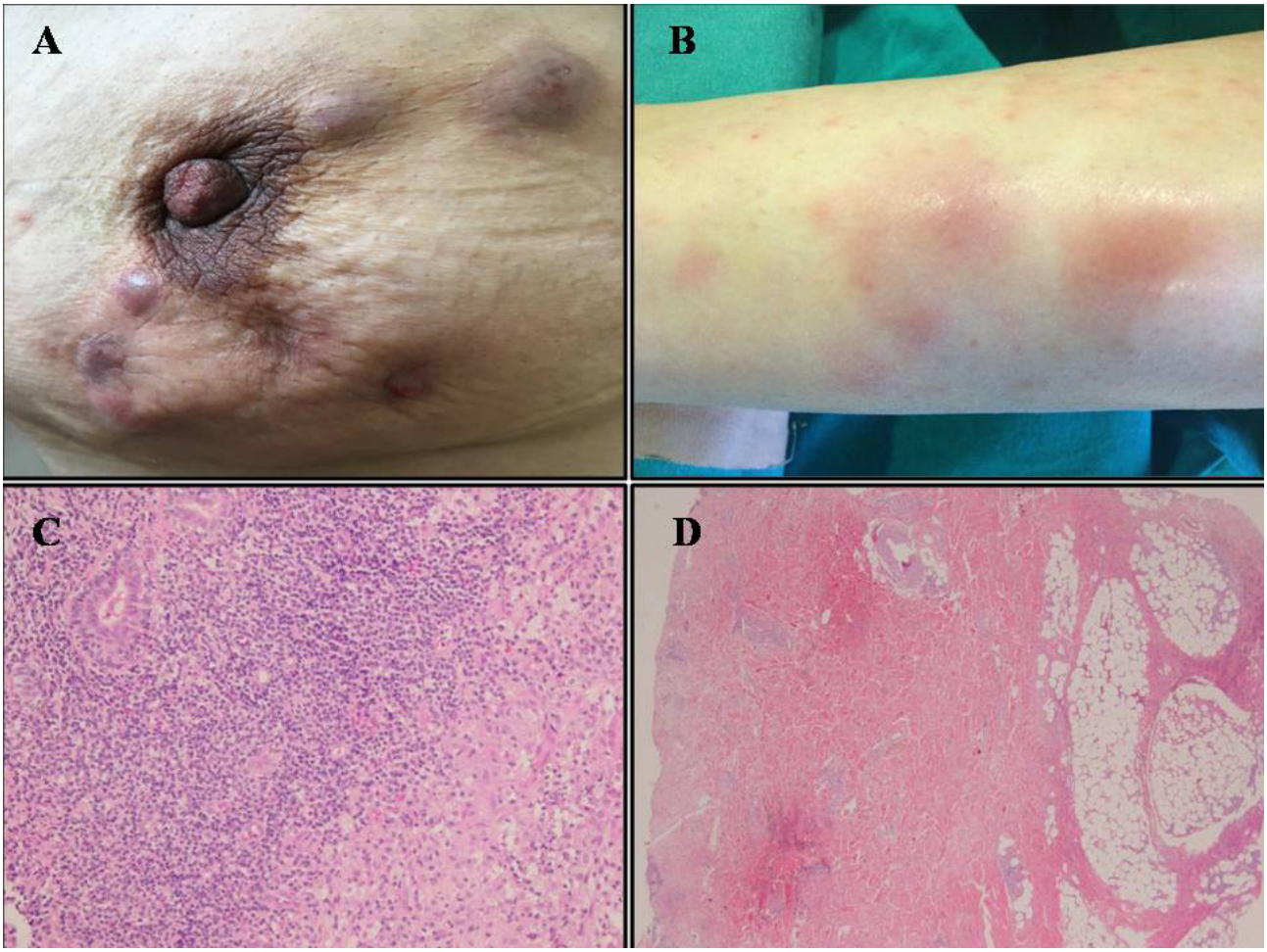

Figure 1A. Granulomatous lobular mastitis on the left breast.
Figure 1B. Erythema nodosum on the left tibia.
Figure 1C. Granulomatous lobular mastitis with characteristic ductolobulocentric pattern (H&E, 200×).
Figure 1D. Erythema nodosum characterized by septal panniculitis in the subcutaneous adipose tissue (H&E, 20×).

chain reaction (PCR) were evaluated when needed for tuberculosis research. Hematoxylin and eosin (H&E) staining for histopathological examination, Gram staining for the detection of microorganisms, Ziehl–Neelsen (ZN) staining for tuberculosis, and periodic acid–Schiff (PAS) staining for fungal infection were applied to all pathological preparations. Serum angiotensin-converting enzyme levels and serum calcium were normal. After radiological examination for breast lesions and dermatological examination for skin lesions, a histopathological examination was done in order to confirm the clinical diagnosis for both lesions (histopathological examination was performed for 4 patients for EN diagnosis). Patients' laboratory results revealed leukocyte counts of >10,000/mm^3 with an elevated erythrocyte sedimentation rate and C-reactive protein titer. Ten patients had EN bilaterally, whereas one patient had a lesion of the right pretibial area. The mean age of the patients was 35.5 years (range: 29–45 years). Four patients had no history of marriage and no reported children. There was history of at least one live birth in the 7 (63.63%) remaining patients. The number of patients with lactation history in the last 12 months was 6 (54.54%), while the number of patients using oral contraceptives was 4 (36.36%). Four patients (36.36%) were smokers. Methylprednisolone was started in the first week at 0.8 mg/kg daily for treatment. The weekly dose was tapered to 0.1 mg/kg daily over 8 weeks.

3. Results

In our series, IGM symptoms and signs had declined and EN lesions had completely disappeared 2 weeks after the start of treatment. Complete recovery of symptoms and signs of IGM occurred after 12 weeks. Patients were followed for an average of 60 (16–110) months after treatment. None of the 11 patients had recurrence or side effects.

4. Discussion

Specific diagnosis of GLM was described for the first time by Kessler and Walloch in 1972 (5). Patients present with a breast mass, nonhealing abscesses, and cutaneous fistulae. In the literature, lesions have been reported to usually affect a single breast, and involvement of the periareolar region has been rarely reported. Redness, thickening of the breast skin, and a palpable hard mass may also be present, as well as some foci of fistula openings in the lesion area (6,7). Our patients also had this type of breast lesion. Diagnosis was confirmed pathologically upon a suspicious mass being detected by radiological evaluation. Histopathologically, noncaseous granulomatous structures consisting mainly of epithelioid histiocytes, lymphocytes, and several Langhans-type giant cells that destruct terminal ductolobular units of the breast with ductolobulocentric patterns can usually be noticed (Figure 1C). On the other hand, histopathologically, EN is characterized by septal panniculitis in the subcutaneous adipose tissue (Figure 1D). Other entities of granulomatous mastitis (tuberculosis, sarcoidosis, fungal infection, etc.) must be studiously excluded. In 1987, Adams et al. described the first case of concurrent IGM and EN (4). Only a few reports are available in the literature presenting several cases (8). Polyarthritis and EN are rare, but they are the most frequent systemic manifestations of IGM. Although cases of IGM complicated solely by EN are very rarely reported, to our knowledge, only ten other cases of IGM complicated by both arthritis and EN have been previously reported. All the patients received immunosuppressive treatment (9,10). It should be considered that IGM and EN may have similar autoimmune mechanisms during their development.

There is still no consensus for IGM treatment in the literature. Corticosteroid therapy was first proposed in 1980 by DeHertog et al. There are studies that advocate the use of steroid treatment at high doses in primary cases, and those advocating that steroid treatment is effective in resistant cases (11). We, as well, started treatment with methylprednisolone, and in all of our cases the initial response to prednisolone was excellent: in 2 weeks the IGM symptoms had markedly declined, while signs of EN had disappeared completely. In our series, patients were followed for an average of 60 months after treatment. None of the 11 patients had recurrence.

In conclusion, we herein report a rare series considering IGM cases complicated by EN. We advocate for an initial trial of methylprednisolone treatment that proved to be very successful in our patients.

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