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CASE REPORT

A Rare Case report of Granulomatous Mastitis with Erythema Nodosum and Polyarthritis which Treat with Topical and Local Steroid

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ABSTRACT

A 36-years-old woman was referred with a painful mass and erythema in the left breast that was initially treated with antibiotics. Three weeks later, the mass and ulceration, erythema had increased and polyarthritis and erythema nodosum had developed in both legs. A core needle biopsy of breast and leg nodule led to a diagnosis of granulomatous mastitis with polyarthritis and erythema nodosum. (GEP) triamcinolon ointment two time for 28 days) and acetaminophen (325 mg two times a day) [clindamycin 150mg three time daily and ciprofloxacin 500 mg two time daily for 15 days]. The size of breast ulceration and mass was decreased. (GEP) are very rare disease and only nine cases have been described, we reviewed the literature to determine the new clinical presentations, (GEP) New appropriate treatment with topical and local steroid injection therapy. And this type of treatment should result in excellent outcomes and prevention complications of the long-time of oral steroid therapy.

CONCLUSION

Early diagnosis and medical treatment of (GEP) is important. Diagnosis should be confirmed by histopathological findings with FNA or core-needle biopsy. Corticosteroid therapy is the choice of treatment for GM. Our patient was started on local and topical steroid.

KEYWORD

Granulomatous mastitis; Erythema nodosum; Polyarthritis; Steroid

INTRODUCTION

Granulomatous Mastitis (GM) is a chronic inflammatory disease of breast characterized by non-caseation granulomas [1]. Although GM is a rare disease of breast, it is very important because it should be to in differential diagnosis of as breast cancer both clinically and radiologically [2]. GM has a chronic course; it begins with a redness and tender mass in breast, proceeding to fistulisation, sinus tract formation, scar formation and deformity of breast if diagnosis and treatment performed with delay and not correctly [2]. In the literatures, there are many theories about the etiology of GM, including autoimmunity, prolactinemia, trauma, alpha-1 antitrypsin deficiency, hormonal and infections; however, these theories have not been proven yet [3]. GM has been rarely associated with oligoarthritis or polyarthritis and erythema nodosum [2,4,5]. Common triggers for EN and polyarthritis include infection, drugs, pregnancy, malignancy, and inflammatory conditions, such as sarcoidosis or gastrointestinal diseases; however, many cases are idiopathic [2,4,6]. The characteristic histologic finding in EN is a septal panniculitis without vasculitis [2,4,6]. EN usually resolves spontaneously within several weeks. When necessary, treatment can be given to reduce symptoms or hasten resolution [2,4,6]. In this case report, we want to discuss the clinical presentations (GEP), diagnostic tools, and new therapeutic approach to a unique case of (GEP) that may be of interest to clinicians or surgeons who are not readily familiar with this condition.

CASE REPORT

A 36-years-old uniparous female patient was admitted to our hospital with one month history of a pain full palpable mass and erythema in left breast (Figure 1 and Figure 2), which was initially treated with antibiotics. Three weeks later, the mass of breast and ulceration, erythema had increased and polyarthritis and erythema nodosum with multiple red tender nodules on both lower extremities and polyarthritis of both knee and ankles, fever and swilling of lower limbs developed (Figure 3). The involved joint was warm and painful. Past medical history of the patient was insignificant. There was history of parturition, breastfeeding, or oral contraceptive drug use in the past three years. Drugs history was negative. Family history for breasts and gynecologic cancer was negative. Breast examination revealed a red tender mass with skin erythema was located on superior portion of left areola in the axillary region there were not any palpable lymph nodes. Ankles of Patient show arthritis of the right and left ankle and both knee with swelling and edema. Widespread erythematous tender nodules on anterior surfaces on both lower extremities were observed (Figure 3). Vital examination for fever was 38 degrees in centigrade. Lab date in Acute phase reactants were elevated [erythrocyte sedimentation rate: 95 mm/hour (normal, 0-20)], [C-reactive protein levels: 119 mg/L (normal, 0-3)]. Leukocyte count was (12800/L). Ultrasound examination of breast excluded breast abscess but showed a 4 cm mass with edema of skin. Cor- needle aspiration biopsy was performed to rule out malignancy (Figure 4 and Figure 5). Tuberculosis work-up was negative. Tuberculosis of breast was excluded with negative of polymerase chain reaction test and acid-fast bacilli staining from aspiration material of the lesion. CXR was normal. Angiotensin converting enzyme (ACE) level was within normal range [14 U/L (normal, 8 U/L - 52 U/L)]. Sarcoidosis and tuberculosis were excluded with these findings and CXR. Antinuclear antibody, anti-neutrophil cytoplasmic antibody was negative and rheumatoid factor level (RF) was within normal range (<6, (normal, 0-14)). Prolactin level was normal (3 ng/mL (normal, 3.3-26.7). ZN stain of needle aspiration for acid fast bacilli was negative. PAS stain was negative for fungal organisms. Gram's stain shows no presence of any bacteria. Culture for mycobacterial tuberculosis yielded no growth. US guided core-needle biopsy was performed. Pathologic examination was compatible with non-necrotizing granulomatous

inflammation consisting of neutrophils, lymphocytes, and multinucleated histiocytic giant cells compatible with granulomatous mastitis (Figure 5). Needle Biopsy of leg lesions revealed Erythema Nodosum. The consultation with pulmonologist and ophthalmologist, they exclude the TB and sarcoidosis, but the rumatologist confirmed the diagnosis of EN and polyartrits. Initial treatment was locally injection of methyprednisolon acetate (40 mg four-week intervals and local triamcinolon ointment two time for 28 days) and acetamenophen (325 mg two times a day). Clindamycin (150 mg three time daily and ciprofloxacin 500 mg two time daily for 15 days). Twenty-eight days after this therapy, Erythema Nodosom and arthritis was regressed (Figure 6). The symptoms and sign of mastitis was regressed partially 50% decreased in size of mass after 28 days during therapy. On follow-up of two months, Erythema Nodosom, and arthritis regressed completely (Figure 7 and Figure 8), and breast mass and erythema subside three months after treatment. Patient was free of symptoms and no relapse was observed through in follow-up of 6 months.

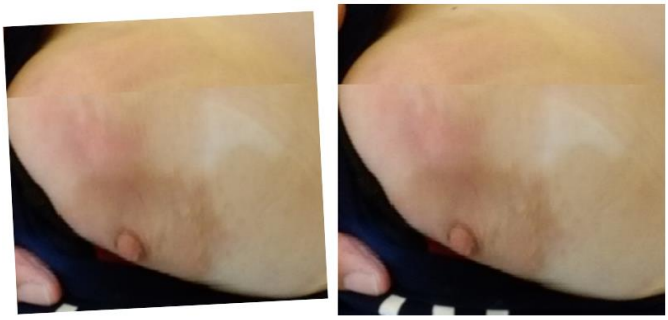


Figure 1 and Figure 2: One month history of a pain full palpable mass and erythema in left breast.

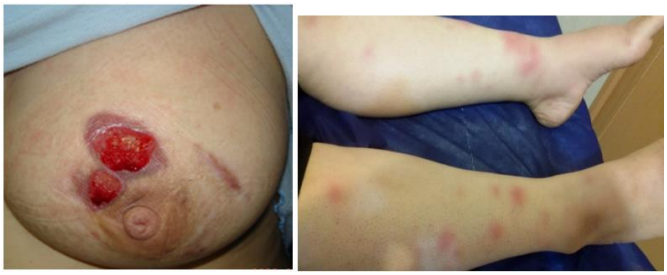


Figure 3 and Figure 4: The mass of breast and ulceration, erythema had increased and polyarthritis and erythema nodosum with multiple red tender nodules on both lower extremities and polyarthritis of both knee and ankles.

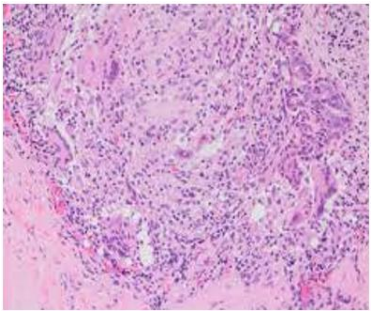


Figure 5: Histopathological examination show aggregates of epithelioid histiocytes, multinucleate giant cells, multinucleated histiocytic giant cells, defined epithelioid granulomas, inflammatory infiltrate consisting of lymphocytes and plasma cells along with granulation tissue fragments.

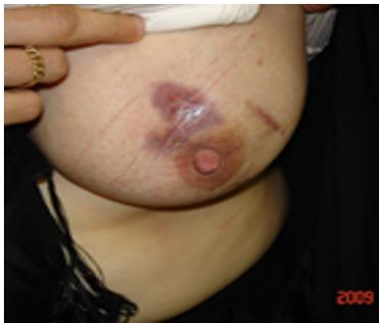
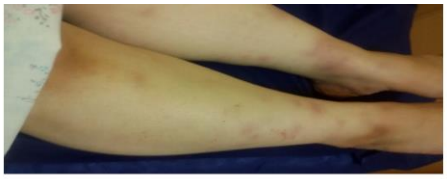


Figure 6: Twenty-eight days after this therapy, Erythema Nodosom and arthritis was regressed.



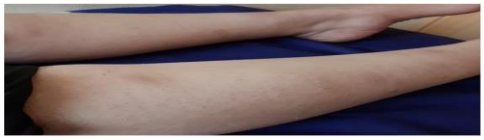


Figure 7 and Figure 8: On follow-up of two months, Erythema Nodosom, and arthritis regressed completely, and breast mass and erythema subside three months after treatment.

DISCUSSION

Granulomatous mastitis (GM) is a rare chronic inflammatory breast disease that can be mistaken with inflammatory or infection and neoplastic disorders of the breast. Erythema nodosum (EN) is a common panniculitis disease. But coexistence of EN and polyartris with GM is very rare [6]. (GM) was first described by Kessler and Wolloch, and is uncommon disease in females, (GM) usually common in reproductive years in female [1,2]. Differentiated diagnosis of GM is infection disease as tuberculosis, bacterial and fungal infection of breast, neoplasms, foreign body reaction, and autoimmune disorders [2-5]. In our case, underlying causes were excluded with medical history, laboratory tests, radiology, and pathology. Many etiological factors including breast feeding, local irritation, oral contraceptive, viral and mycotic infections, parasites disease, hyperprolactinemia, diabetes mellitus, smoking, alpha-1 antitrypsin deficiency, and autoimmunity have been suggested in pathogenesis of GM, but none of them have been proven yet [2,5]. Erythema nodosum (EN) is an acute, nodular, erythematous eruption that usually is limited to the extensor aspects of the lower legs [6,7]. Chronic or recurrent erythema nodosum is rare but may occur [7,9,10]. Erythema nodosum is presumed to be a hypersensitivity reaction and may occur in association with several systemic diseases or drug therapies, or it may be idiopathic [6,7,10]. The inflammatory reaction occurs in the panniculus form [2,4,5]. Polyarthritis and erythema nodosum are rare systemic manifestations of granulomatous mastitis [8,10]. In 1987, Adams and colleagues reported the first case of granulomatous mastitis coextended with arthritis and erythema nodosum [7]. Only ten other cases of granulomatous mastitis coextend by both arthritis and erythema nodosum have been previously reported [6,7,10]. Response of GM to steroids and methotrexate therapy and some disease as erythema nodosum or arthritis, support an autoimmune process in the etiology GM [9,10]. Usually, GM presents as unilateral breast mass, breast pain, nipple inversion, skin retraction and ulceration with axillary lymphadenopathy [6,7,10]. Biopsy site of breast with

core-needle or open biopsy usually fail to repair for long time [5]. Biopsy material from the center of lesion is valuable for histopathological examination for accurate diagnosis [2,5,8]. Pathological findings in GM are no castings Granuloma formations, dense neutrophilic infiltration without necrosis [6-8]. Other inflammation disease of breast as tuberculosis, actinomyces and systemic diseases such as sarcoidosis, Wegener granulomatosis should be excluded [6,8]. Taylor detected Corynebacterium in 41% of 34 isolated GM lesions [8]. In the literature, association of EN, GM, and polyarthritis has been reported in seven cases up until now [4,9]. Two of these reports were happened in pregnancy duration and in third trimester [10]. Resolution time in some cases may prolong up to 9 months to 12 months [2,4,5,8]. Moderate and severe disease of GM require systemic treatment [11-13]. Surgery is not recommended due to slow wound healing after the operation [5,10]. Prednisolon is the drug of choice for treatment of GM [5,10,12]. Methotrexate is reported as an effective treatment option in a few studies' colchicine, hydroxychloroquine and systemic steroids were reported to be effective treatment modalities [5,10,12-14]. We used local and topical injection of methylprednisoln acetate for treatment in our patient. Despite therapy, recurrences may be occurring [5,10]. (GEP) in our case was unresponsive to NSAD and others anti-inflammatory drugs or antibiotic in other clinic before admission to our clinic and because of complication of oral steroid therapy, we used local and topical steroid in our patient and response was very good to local and topical injection therapy after 8 weeks. And in six-month follow-up recurrence was not happened.

CONCLUSION

Early diagnosis and medical treatment of MG and EN is important. Diagnosis should be confirmed by histopathological findings with core-needle biopsy. Corticosteroid therapy is the choice of treatment for GM. Our patient was started on local and topical steroids and had marked improvement in her symptoms and reduction in the size of breast lump. Surgery not indicated in GM. Early diagnosis and medical treatment may prevent patients from undergoing potentially disfiguring surgery.

REFERENCES

1. Kessler E and Wolloch Y (1972) Granulomatous mastitis: A lesion clinically simulating carcinoma. American Journal of Clinical Pathology 58(6): 642-646.
2. Patel RA, Strickland P, Sankara IR et al. (2010) Idiopathic granulomatous mastitis: Case reports and review of literature. Journal of General Internal Medicine 25(3): 270-273.
3. Pereira FA, Mudgil AV, Macias ES et al. (2012) Idiopathic granulomatous lobular mastitis. International Journal of Dermatology 51(2): 142-151.
4. Zabetian S, Friedman BJ, McHargue C (2016) A case of idiopathic granulomatous mastitis associated with erythema nodosum, arthritis, and reactive cough. JAAD Case Reports 2(2): 125-127.
5. Aghajanzadeh M, Hassanzadeh R, Sefat SA et al. (2015) Granulomatous mastitis: Presentations, diagnosis, treatment and outcome in 206 patients from the north of Iran. The Breast 24(4): 456-460.
6. Hida T, Minami M, Kawaguchi H et al. (2014) Case of erythema nodosum associated with granulomatous mastitis probably due to *Corynebacterium* infection. The Journal of Dermatology 41(9): 821-823.
7. Adams DH, Hubscher SG, Scott DG (1987) Granulomatous mastitis--a rare cause of erythema nodosum. Postgraduate Medical Journal 63(741): 581-582.
8. Nakamura Y, Yamaguchi M, Nakamura A et al. (2014) Tender, red nodules and arthralgia in a young woman with mastitis. Clinical and Experimental Dermatology 39(3): 410-412.

9. Taylor GB, Paviour SD, Musaad S et al. (2003) A clinicopathological review of 34 cases of inflammatory breast disease showing an association between corynebacteria infection and granulomatous mastitis. Pathology 35(2): 109-119.
10. Olfatbakhsh A, Beheshtian T, Djavid GE (2008) Granulomatous mastitis, erythema nodosum, and oligoarthritis in a pregnant woman. The Breast Journal 14(6): 588-590.
11. Binesh F, Shiryazdi M, Owlia MB et al. (2013) Idiopathic granulomatous mastitis, erythema nodosum and bilateral ankle arthritis in an Iranian woman. Case Reports 2013: bcr2012007636.
12. Al-Khaffaf B, Knox F, Bundred NJ (2008) Idiopathic granulomatous mastitis: A 25-year experience. Journal of the American College of Surgeons 206(2): 269-273.
13. Kornfeld HW and Mitchell KB (2021) Management of idiopathic granulomatous mastitis in lactation: Case report and review of the literature. International Breastfeeding Journal 16(1): 1-6.
14. Ertürk TF, Çakır Ö, Yaprak Bayrak B et al. (2021) Local steroid treatment: An effective procedure for idiopathic granulomatous mastitis, including complicated cases. Journal of Investigative Surgery: 1-7.