Idiopathic Granulomatous Mastitis, Erythema Nodosum, and Polyarthritis

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ABSTRACT: Idiopathic granulomatous mastitis (IGM) is an inflammatory-mediated rare disease that can be linked to rare manifestations. Erythema nodosum (EN) and polyarthritis, seen in a multitude of autoinflammatory and autoimmune diseases, have been rarely linked to IGM. Despite the cause of IGM being unclear, Corynebacterium infections are thought to play a role in the pathophysiology of IGM. Unusually, IGM has a relapsing and remitting course, which also applies to its systemic manifestations. As such, we present a case of IGM in a middle-aged lady who was initially thought to have Corynebacterium-containing unilateral abscesses for which drainage was performed. However, several abscesses devoid of bacterial growth started recurring, and the disease course was complicated by EN and polyarthritis. IGM, EN, and polyarthritis eventually resolved and were managed with symptomatic treatment.

KEYWORDS: Idiopathic granulomatous mastitis, erythema nodosum, polyarthritis, Corynebacterium

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Introduction

Idiopathic granulomatous mastitis (IGM) is a rare benign inflammatory disease of the breast that can mimic other breast pathologies.[1] Despite that the cause of IGM is not well understood, multiple risk factors were described, most importantly including recent pregnancy and/or breastfeeding.[2]

The presentation consists of unilateral or bilateral tender firm breast masses accompanied by erythema, pain, and drainage.[3] The diagnosis is ideally performed by breast biopsy (ideally by core biopsy) showing a noncaseating granuloma with epithelioid histiocytes and multinucleated giant cells within breast lobules, and might contain micro abscesses.[2]

Certain diseases have been associated with IGM, such as tuberculosis or sarcoidosis, and less rarely ANCA-associated vasculitis.[4] Certain manifestations, that are linked with rheumatological conditions, have been described in the context of IGM. Erythema nodosum (EN) and polyarthritis are examples of such manifestations. We present a case of a middle aged woman who had relapsing and remitting unilateral breast abscesses to be diagnosed with IGM followed by appearance of EN and polyarthritis, both of which self-resolved with resolution of mastitis lesions.

Case Presentation

A 41-year-old G5 P5 previously healthy female presented with vague pain, discharge, and erythema in her right breast. The patient's last lactation occurred approximately 3 years before her current presentation. Despite attempting self-medication with metronidazole, the symptoms recurred after discontinuing the antibiotic.

Mammography and breast ultrasound revealed a 7-cm nodular and irregular area involving the right breast at 8 o'clock (Figure 1A and B). In addition, there was a suspicious right axillary lymph node 21 mm (Figure 1C and D). However, no abnormalities were detected in the left breast. Biopsy of the breast mass showed widespread acute inflammation, abscess formation, and fat necrosis (Figure 2).

As there was no clinical improvement with pharmacological therapy, mastotomy with exploration and abscess drainage was performed, and tissue culture revealed the growth of pan susceptible Corynebacterium species. Amoxicillin-clavulanic acid was administered for a 7-day course.

For approximately 1 year after the operation, the patient experienced no symptoms. However, she later developed similar symptoms in the same breast, with new areas being involved on clinical examination. Incision and drainage were performed, but no growth of bacteria was observed.

A few months later, new abscesses began to appear at distinct intervals, all of which resolved completely with short courses of non-steroidal anti-inflammatory drugs (NSAIDs). The patient experienced relief from severe pain when the abscesses self-drained. Notably, the appearance of abscesses was accompanied by flares of fever, myalgias, and polyarthritis, involving the right ankle, hands, and feet, and erythematous and tender bumps were noticed symmetrically on the shins (Figure 3), diagnosed as erythema nodosum. However, the

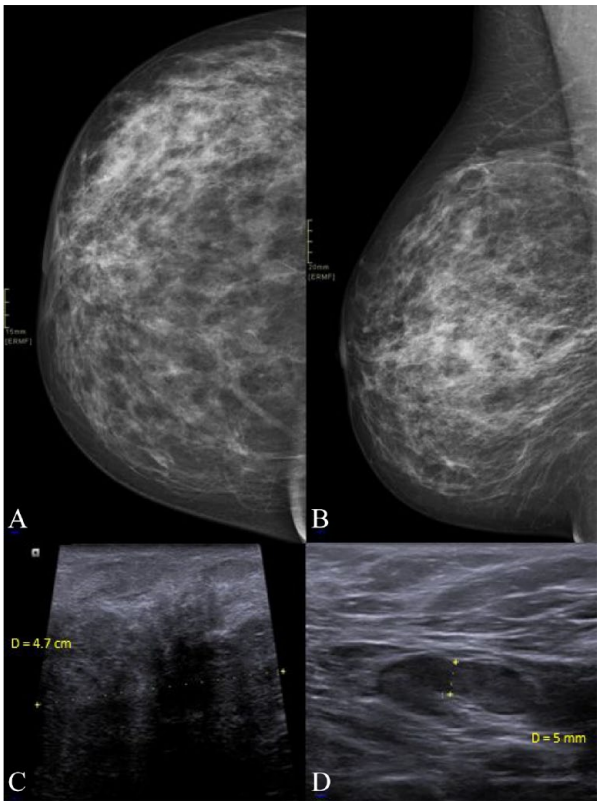

Figure 1. Craniocaudal (A) and mediolateral oblique views (B) of the right breast show an area of increased density along the outer slightly lower quadrant of the right breast. Ultrasound (C) of this region showed an area of decreased echogenicity with areas of low-level internal echoes. Aspiration of the areas with low-level internal echoes revealed pus, which was sent for cytology and culture. Ultrasound of the right axillary region (D) showed a borderline 2 × 1 cm lymph node with 5 mm cortical thickening.

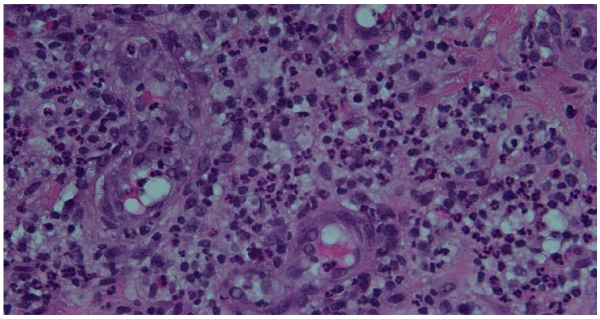

Figure 2. Acute inflammation consisting of neutrophils with prominent vessels. Scattered eosinophils and lymphocytes are also seen. (Hematoxylin and Eosin stain, original magnification 40×).

polyarthritis and erythema nodosum resolved with the resolution of abscesses.

Discussion

Idiopathic granulomatous mastitis (IGM) is a benign rare condition which involves women of childbearing age.[5] The true prevalence of IGM is unknown. The course of the disease is characterized by a chronic relapsing and remitting inflammation, resulting in scarring and discharging sinuses.[6] Multiple causes were suggested for IGM, although the disease pathophysiology remains unclear. Examples of causes include autoimmune diseases, hormone irregularities, and local immune response to trauma.[1] Nevertheless, sarcoidosis, tuberculosis, foreign substances and bacterial infections are considered to be causes.[1] Specifically, Corynebacterium has been described as a significant contributor to the development of IGM and a cause for IGM recurrence.[7]

Polyarthritis has been reported as a rare complication of IGM. Polyarthritis can occur alone or might be associated with other complications such as erythema nodosum (EN).[8] Corynebacterium has been rarely linked to erythema nodosum.[9] Interestingly, this association has happened in the context of IGM. Furthermore, Corynebacterium has been linked to arthritis, specifically septic arthritis.[10]

Table 1 lists literature cases that linked IGM with EN and polyarthritis. Interestingly, most cases were treated with corticosteroids. Only 2 previous cases' manifestations resolved spontaneously[26] similar to our case, and only 1 case where IGM, polyarthritis, and EN coexist in the setting of a Corynebacterium infection.[25]

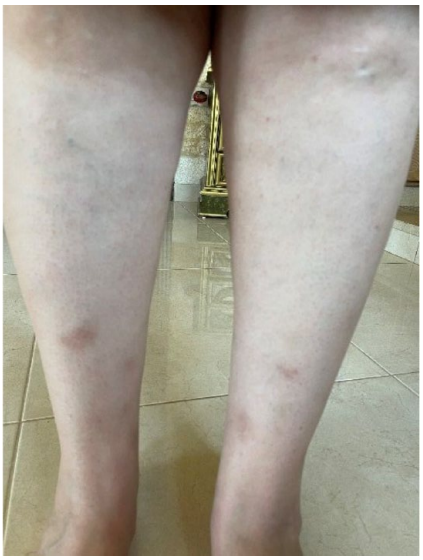

Figure 3. Raised erythematous spots on the anterior shins bilaterally.

Conclusion

Idiopathic granulomatous mastitis can be preceded by a Corynebacterium breast abscess resulting in relapsing and remitting unilateral breast involvement. Erythema nodosum and polyarthritis are 2 complications that can follow the same disease course of IGM. Although glucocorticoids and methotrexate have been described in the literature, our experience shows that conservative and symptomatic management can be sufficient for resolution of symptoms, although a close follow-up is needed to assess for recurrence. The choice of therapy depends on the frequency, duration, and severity of the episodes, response to symptomatic management, and the symptoms overall effect on patients' quality of life.

Table 1. Previous literature cases linking idiopathic granulomatous mastitis to erythema nodosum and polyarthritis.

REFERENCE	AGE	MEDICAL HISTORY	RACE	MANAGEMENT
Binesh et al[11]	40	None	Middle Eastern (Iranian)	Dexamethasone followed by prednisolone
Zabetian et al[12]	43	None	Hispanic	Prednisone followed by azathioprine
Şener Bahçe and Aktaş[13]	20	Pregnant First trimester	Not mentioned	Prednisolone
	25	Pregnant Third trimester	Not mentioned	Prednisolone
	29	Not mentioned	Not mentioned	Prednisolone
	37	Pregnant Third trimester	Not mentioned	No treatment
	30	Pregnant Third trimester	Not mentioned	Prednisolone
	41	Not mentioned	Not mentioned	Prednisolone
Adams et al[14]	24	Not mentioned	Not mentioned	Indomethacin followed by rifampicin and isoniazid
Vural et al[15]	32	None	Turkish	Indomethacin and potassium iodide solution followed by colchicine
Salesi et al[16]	23	Pregnant Third trimester	Iranian	Prednisolone and colchicine and azathioprine
Laor et al[17]	16	None	Hispanic	Wide local excision followed by trimethoprim-sulfamethoxazole followed by naproxen and prednisone.
Parperis et al[18]	40	None	Caucasian	Prednisone and antibiotics and NSAIDs
	38	None	Caucasian	Prednisone and antibiotics and NSAIDs
Akao et al[19]	27	None	Not mentioned	Prednisolone
Alungal et al[20]	25	None	Indian	Oral corticosteroids
Cammarata et al[21]	31	None	Turkish	Prednisone
Ben Abid et al[22]	36	None	Indian	ATT and NSAIDs
Olfatbakhsh et al[23]	30	Pregnant Third trimester	Iranian	Low-dose corticosteroids
Haddad et al[24]	Not mentioned	Pregnant Second trimester	Iranian	No treatment
Yoshino et al[25]	37	Pregnant Second trimester	Not mentioned	Drainage and prednisolone

Abbreviations: ATT, anti-tuberculous treatment; NSAIDs, Non-steroidal anti-inflammatory drugs.

Ethics approval

Ethical permission was not required since this is a case report.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Author contributions

Ali Tarhini: Investigation; Writing—original draft
Georges El Hasbani: Formal analysis; Writing—original draft; Writing—review & editing
Lama Farhat: Formal analysis
Diamond Ghieh: Conceptualization; Writing—original draft
Imad Uthman: Writing—review & editing

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Competing interests

The authors declare that there is no conflict of interest.

Availability of data and materials

None.

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