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A very rare case of eosinophilic mastitis

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ABSTRACT

INTRODUCTION: Eosinophilic mastitis caused by eosinophil infiltration of the mammary gland is very rare. To date, no report has been published on treating patients with this disorder using anti-allergic drugs. Steroids are commonly used in these cases, but have greater burden.

PRESENTATION OF CASE: A 33-year-old woman presented to the author's clinic with a tumor and pain in the upper inner quadrant of the left breast. She underwent core needle biopsy (CNB) and was diagnosed with eosinophilic mastitis based on histopathological analysis. The serum eosinophil count at the time of biopsy increased to 1560/μL. She was administered 100 mg of suplatast tosilate (brand name: IPD capsule 100), an anti-allergic drug, 3 times daily after each meal.

Thereafter, the patient's symptoms improved and her serum eosinophil count returned to normal after 4 months. To date, the patient has been recurrence-free for 3 years since the first presentation.

DISCUSSION: Organ damage induced by eosinophil infiltration was limited to the mammary gland and improved with anti-allergic drug administration.

CONCLUSION: This report presents the successful treatment of an isolated case of eosinophilic mastitis solely using anti-allergic drugs.

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1. Introduction

Eosinophil infiltration of the mammary gland is rare. Very few cases of eosinophilic mastitis have been reported, and most have been treated with steroidal agents. Eosinophilic mastitis with organ damage limited to the mammary gland is even rarer.

To date, no report has described the successful treatment of a patient with eosinophilic mastitis using only anti-allergic drugs. Herein, a case of mastitis caused by eosinophilic infiltration successfully treated with anti-allergic drugs is presented. This report conforms to the SCARE criteria [1].

2. Case report

A 33-year-old woman detected a tumor and pain in the upper-inner quadrant of the left breast 1 week before her visit to the author's Clinic. Her medical history showed that she had asthma from the age of 18 months to 15 years. However, she was cured and showed no asthma symptoms thereafter. Her family history was unremarkable. Mammography showed a localized asymmetric nodular shadow in the craniocaudal view of the left breast, and she was diagnosed with category 3 (Fig. 1a, b). Ultrasonography showed a heterogeneous tumor of 41.3 × 38.0 × 24.0 mm in the upper inner quadrant of the left breast (Fig. 2a). The border and margin of the tumor were unclear; it has no halo or change in the posterior echoes. Color Doppler of ultrasonography showed a nearly spotty and plunging vascular pattern (Fig. 2b, c).

She underwent CNB for the mammary gland tumor to rule out malignant disease and to get a diagnosis of the tumor. As a result, she was diagnosed with mastitis with prominent eosinophilia, consistent with eosinophilic mastitis. It was entirely fibrotic and accompanied by severe diffuse infiltration of eosinophils and lymphocytes. Glandular epithelial cells were slightly hyperplastic but showed no other marked change. Biopsy specimen did not show distinct vasculitis (Fig. 3a, b). The blood test showed that the peripheral eosinophil level increased to 1.560/μL. Computed tomography examination showed a weak, ill-defined, diffusive enhancement in the upper inner left breast. The site was slightly swelled but showed no distinct tumor formation. The bilateral lung fields were normal, and no hepatosplenomegaly was observed (Fig. 4a, b). Magnetic resonance imaging examination showed no image of apparent tumor in the bilateral breasts including the upper inner left breast (Fig. 5a–c).

The eosinophil level was 1560/μL before treatment and decreased to 1121/μL 2 weeks after administration of anti-allergic agents, which further improved to 732/μL after 1 month and

Abbreviations: HES, hypereosinophilic syndrome; CNB, core needle biopsy; CSS, Churg-Strauss syndrome.

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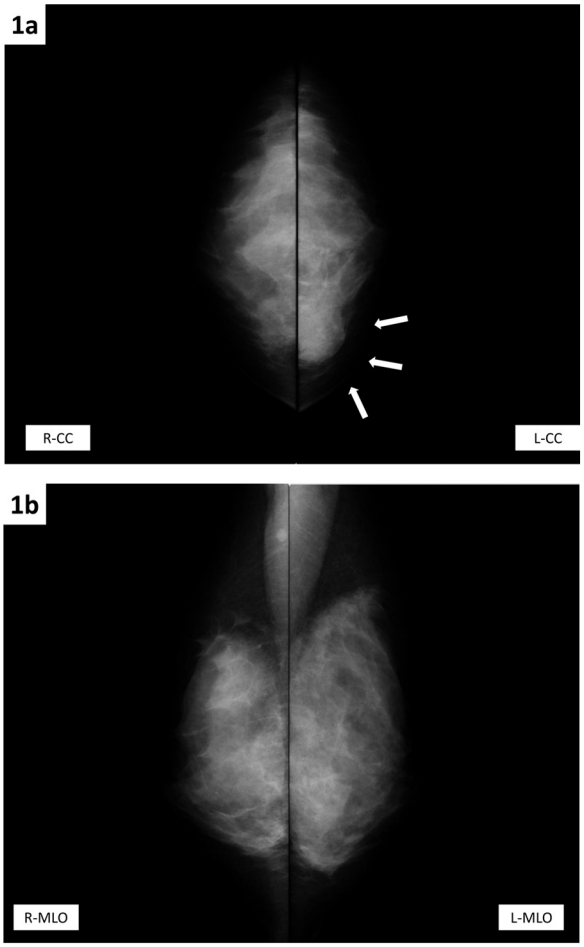


Fig. 1. Mammography; 1(a,b). Mammography showed a localized asymmetric nodular shadow in the craniocaudal view of the left breast, and she was diagnosed with category 3 cancer.

385/μL after 4 months. The induration and pain in the upper inner left breast improved along with improvement in the serum eosinophil level. No symptom recurrence has been reported during the annual follow-up three years later.

3. Discussion

Very few cases of eosinophilic mastitis have been previously reported [2–5]. Patients are diagnosed with eosinophilia if their serum eosinophil count is greater than 450/μL. In the case presented here, the blood test conducted before treatment showed that the serum eosinophil count increased to 1560/μL.

CNB confirmed the diagnosis of eosinophilia. Owing to the good general condition of the patient, steroid administration was considered. However, after confirming that the use of the light anti-allergic drugs had a lower burden than the use of steroids, the patient was administered anti-allergic drugs immediately after the diagnosis was confirmed, following which she gradually recovered.

Hypereosinophilic syndrome (HES) results from eosinophilia of unknown origin lasting for at least 6 months; eosinophil infiltration into organs consequently leads to organ disorders.

The diagnostic criteria for HES include eosinophilia of unknown origin; serum eosinophil count of at least 1500/μL for 6 months or more; no distinctive cause of eosinophilia such as allergic or parasitic disease and malignant tumors; and organ disorder caused by eosinophil infiltration [6–8].

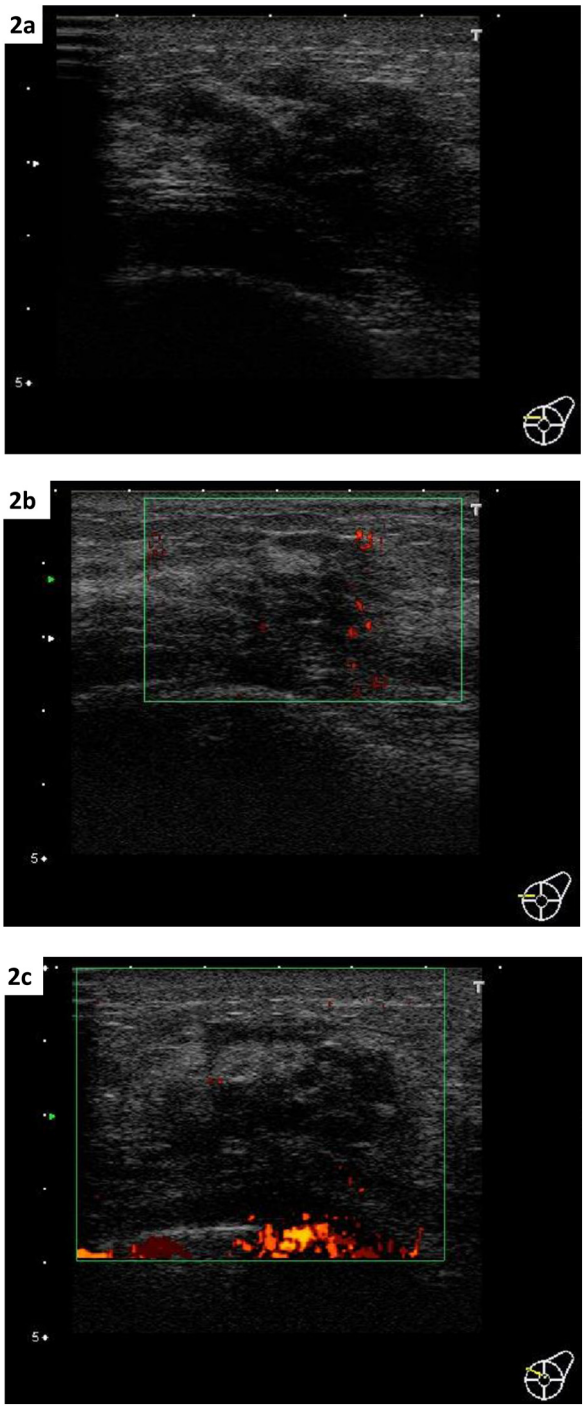
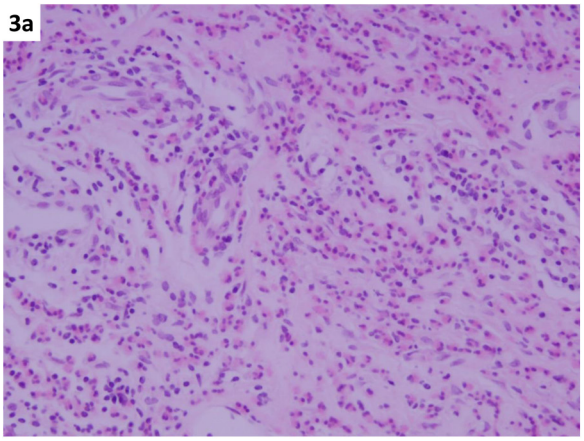


Fig. 2. Breast ultrasonography; (2a) Ultrasonography showed a heterogeneous tumor of 41.3 × 38.0 × 24.0 mm in the upper inner quadrant of the left breast. The border and margin of the tumor were unclear; it has no halo or change in the posterior echoes. (2b, c) Color Doppler of ultrasonography showed a nearly spotty and plunging vascular pattern.

In this case, organ damage was induced by eosinophil infiltration in the mammary gland. The patient had no parasitic or allergic disease at the time of onset, such as bronchial asthma, which had been observed in her infancy. Computed tomography and clinical findings excluded malignant tumors.

The mechanism of organ disorders associated with eosinophils has previously been described [9,10]. In brief, eosinophil activation causes degranulation and release of cytotoxic proteins such



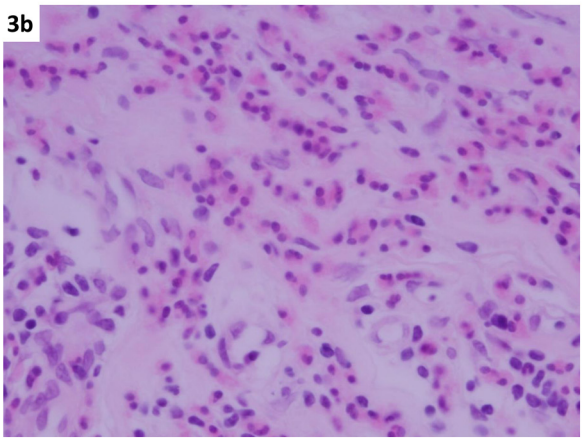


Fig. 3. Pathological analysis; 3(a, b). Analysis of the CNB specimen.
Core needle biopsy (CNB) for the mammary gland tumor and was diagnosed with mastitis with prominent eosinophilia, consistent with eosinophilic mastitis. It was entirely fibrotic and accompanied by severe diffuse infiltration of eosinophils and lymphocytes. Glandular epithelial cells were slightly hyperplastic but showed no other marked change. Biopsy specimen did not show distinct vasculitis. (3a: [H-Estain, ×50], 3b:[H-Estain, ×100]).

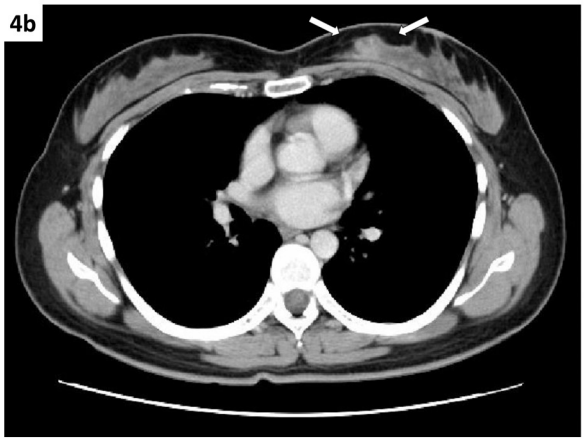


Fig. 4. Computed tomoguraphy; 4(a, b). Computed tomography examination showed a weak, ill-defined, diffusive enhancement in the upper inner left breast. The site was slightly swelled but showed no distinct tumor formation. The bilateral lung fields were normal, and no hepatosplenomegaly was observed.

as major basic protein, eosinophilic cationic protein, eosinophil-derived neurotoxin, and eosinophilic peroxidase.

Patients with HES are commonly treated with steroidal agents to suppress eosinophil infiltration and degranulation. In this case, anti-allergic drugs were administered instead of steroidal agents, because the patient had organ damage limited to the mammary gland and anti-allergic drugs have a lower burden than steroidal drugs. Treatment with anti-allergic drugs was continued while symptoms and eosinophil count improved. No recurrence occurred after the discontinuation of the drugs.

The patient noticed her symptoms 1 week before her first visit to the author's clinic. She did not report of having these symptoms before that time. In addition, the patient's symptoms were not serious compared with those of patients with HES, which is why anti-allergic drugs were used instead of steroidal agents. From these observations, there is no apparent evidence that this patient might have HES.

Another disorder, Churg-Strauss Syndrome (CSS) is secondary to bronchial asthma and/or allergic rhinitis and it increases serum eosinophils and causes tissue damage most commonly in the lungs and the digestive tract. It is an autoimmune condition that causes inflammation of small- and medium-sized blood vessels. CSS also causes granulomatous angiitis and necrotizing systemic angiitis. Extravascular granulomatosis has been associated with hypereosinophilia as well as eosinophilic infiltration inside and outside of blood vessels, which can eventually lead to cell death and be life-threatening.

CSS is also known as eosinophilic granulomatosis with polyangiitis or allergic granulomatosis angiitis [11–13]. Effective treatment of patients with CSS requires suppression of the immune system with medication, including steroidal agents and cyclophosphamide or azathioprine. In this case, the histopathological analysis of the CNB of the mammary gland did not show findings suggestive of vasculitis. It is less likely that the patient had CSS.

This is an isolated case report describing the successful treatment of a patient with mastitis due to eosinophil infiltration by using anti-allergic drugs.

4. Conclusion

This case report presents the diagnosis of a very rare case of eosinophilic mastitis, which improved with the administration of anti-allergic drugs. The serum eosinophil count also returned to normal. No recurrence was observed after the discontinuation of anti-allergic drugs.

This report demonstrated the successful treatment of a patient with an isolated case of mastitis due to eosinophil infiltration using anti-allergic drugs. This outcome suggests that anti-allergic drugs may be superior to steroids as a first-line treatment in patients with eosinophilic mastitis and organ damage to the mammary gland.

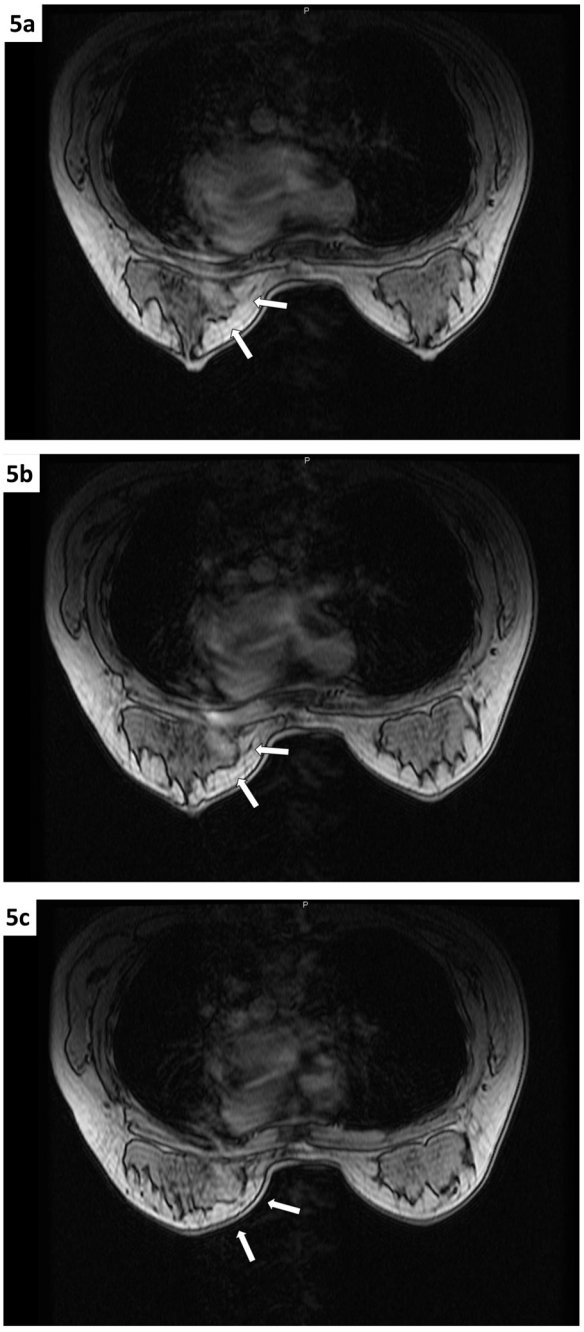


Fig. 5. Magnetic resonance imaging; 5(a–c). Magnetic resonance imaging examination showed no image of apparent tumor in the bilateral breasts including the upper inner left breast.

Conflict of interest

No conflict of interest.

Funding

No funding.

Ethical approval

IRB/Ethics Committee ruled that approval was not required for this study.

Consent

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

Author contribution

Keiichi Takahashi is the sole author.

Registration of research studies

Not applicable.

Guarantor

Keiichi Takahashi.

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