



A Rare Case of Granulomatous Mastitis in the Accessory Axillary Breast of a Pregnant Woman Successfully Treated by Surgery

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ABSTRACT

Granulomatous mastitis (GM) is a chronic inflammatory disease of the breast that usually occurs in women of reproductive age. However, GM during pregnancy is unusual and only one case of GM in the accessory breast has been reported so far. Here, we report an extremely rare case of GM in the accessory axillary breast of a pregnant woman. A 24-year-old pregnant woman had persistent pain and swelling in the right axilla that did not improve with antibiotic administration. Despite incision and drainage for subcutaneous abscess, the incised skin gradually became ulcerated, exposing the subcutaneous granulomatous tissue. *Corynebacterium* species were isolated in the bacterial culture of drained pus. Lower back pain, pain in several joints, and erythema nodosum on the lower legs appeared later. Based on the result of bacterial culture and the above disease course, the patient was clinically suspected of having GM. The axillary mass was surgically removed after childbirth, and the excised mass was histopathologically confirmed as GM. Treatment for GM should be considered individually and carefully in accordance with the patients' condition. Unnecessary surgery should be avoided. However, early addition of surgical interventions may yield good outcomes, especially for pregnant women because of limited treatment options.

Keywords: Axilla; granulomatous mastitis; pregnancy; surgery

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Key Points

- Granulomatous mastitis (GM) usually occurs in women of reproductive age, and only one case involving the accessory breast has been reported.
- No standard management for GM has been established, so treatment strategy should be planned individually.
- In the present case, surgery provided an early recovery from GM in the axilla during pregnancy.

Introduction

Granulomatous mastitis (GM) is an uncommon chronic inflammatory disease of the breast that presents with symptoms such as breast mass, abscess, erythema, induration, and tenderness. Although the etiology underlying GM remains unclear, a localized autoimmune inflammatory response to milk in the duct has been implicated in its pathogenesis. Thus, a correlation with breastfeeding and childbirth has been investigated (1). Previous reports have also associated GM with other clinical manifestations, such as erythema nodosum and, occasionally, with arthritis, suggesting that GM has an autoimmune component (2). Paviour et al. (3) was the first group to isolate *Corynebacterium* species in nine of 12 cases of GM. *Corynebacterium* is a lipophilic, Gram-positive, rod-shaped bacterium. Due to the lipophilicity, *Corynebacterium* infection has recently been suggested to be associated with the development of GM in lipid-rich mammary glands (3). Therapeutic strategies for GM include simple observation, antibiotic administration, steroid administration, drainage, excision, mastectomy, and combinations thereof. Here, we report a case of GM in the accessory axillary breast of a pregnant woman successfully treated by surgery.

Case Presentation

A 24-year-old pregnant woman visited an obstetrics clinic at 28 weeks 3 days of gestation with complaints of pain and swelling in the right axilla that had persisted for one week. Although intravenous piperacillin was given for three days, the symptoms worsened. Therefore, the patient was referred to our hospital. A fist-sized mass with redness and heat was noted in the right axilla (Figure 1). Ultrasonography revealed subcutaneous

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abscess formation, so incision and drainage were performed on suspicion of pyogenic mastitis. However, the incised skin gradually became ulcerated, and the granulomatous tissue was exposed (Figure 2). *Corynebacterium* species were later isolated in the bacterial culture of drained pus. Pain in the lower back, elbows, hands, and ankles appeared at 30 weeks 5 days of gestation, and erythema nodosum appeared on the lower legs at 31 weeks 3 days of gestation. Based on the result of bacterial culture and the natural history of the condition, the patient was clinically suspected of having GM. A needle biopsy was considered to achieve definitive diagnosis, followed by systemic steroid therapy. However, the patient opted for elective surgery after childbirth, because she was about to give birth and had already endured a long period of suffering. Although two weeks of hospitalization was required from 34 weeks 3 days of gestation because of imminent premature birth, the patient gave birth at 36 weeks 3 days of gestation. The axial mass was surgically removed one week after delivery (Figure 3), and the excised mass was histopathologically confirmed as GM (Figure 4).

Discussion and Conclusion

Accessory breast tissue is subject to the same diseases as normally located breast tissue. The most frequent diseases reported in the accessory breasts are cancers, followed by mastitis, fibroadenoma, phyllodes tumor, and fibrocystic change (4). Yilmaz et al. (5) have recently reported a case of GM involving the accessory axillary breast. However, to our knowledge, there have been no other reports of GM in accessory breast tissue. GM usually occurs in women of reproductive age, and most cases occur around two years after breastfeeding, while GM during pregnancy is unusual (1). Moreover, GM is known to cause systemic inflammatory reactions, such as erythema nodosum and arthritis, as seen in our patient (1). It has been shown that rheumatologic conditions were present in 34% of published cases and erythema nodosum in 8% (6).

Although a standard therapeutic strategy has not yet been established, steroids are often administered for GM (6). Steroid therapy for GM was first described by DeHertogh et al. (7) in 1980, who recommended 30 mg/day prednisone for at least two months. Nevertheless, no



Figure 1. An image of the patient at the first visit
Arrow: accessory nipple



Figure 2. An image of the patient at 36 weeks 3 days of gestation. Incised skin was ulcerated and the granulomatous tissue was exposed



Figure 3. Images of the patient's right axilla (left) and breasts (right) 40 days after surgery

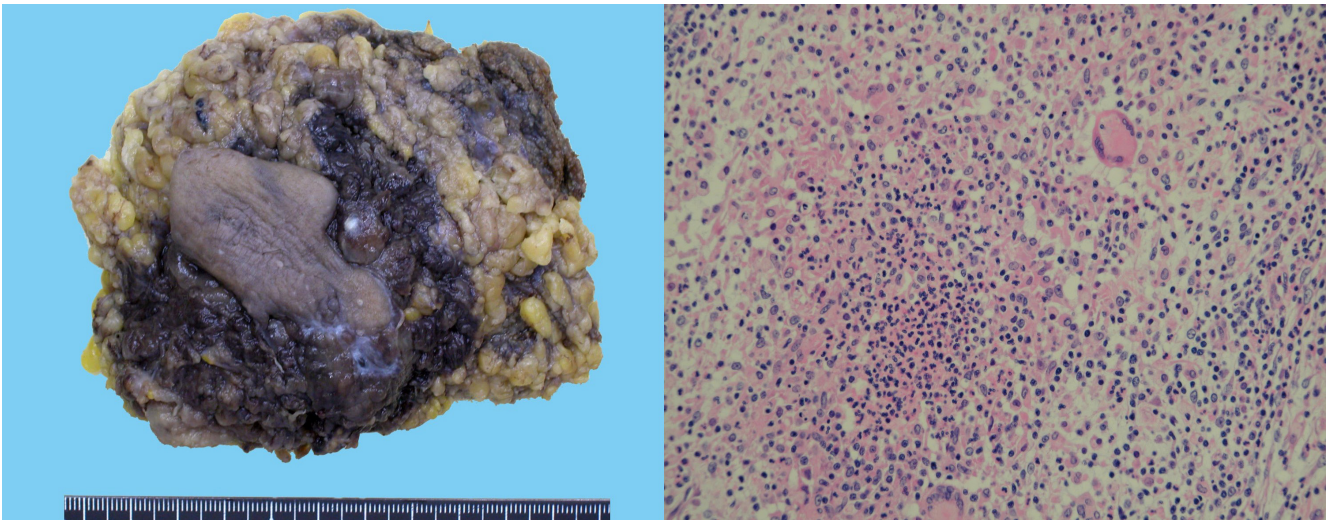


Figure 4. Excised mass (left) and pathological examination (right) showing granulation tissue with marked inflammatory cell infiltration, including multinuclear giant cells and abscess formation

consensus has been reached on the optimal dose and duration of treatment. This intervention can lead to a decrease in the diameter of the lesion, while adverse effects, such as Cushing syndrome, weight gain, hyperglycemia (1), and opportunistic infections are possible (8). Moreover, steroid treatment of GM during pregnancy has not been addressed to date. A few cases of GM during pregnancy were successfully treated with steroids (9, 10) but no information on adverse effects of the treatment and outcome of pregnancy were provided. Furthermore, it is suggested that exposure to steroids during early pregnancy increases the risk of oral cleft, premature delivery, and low birth weight (11, 12). Systemic steroid treatment and/or intralesional and topical steroid applications may have been alternatives in the present case, because our patient was in late pregnancy. However, in patients who are positive for *Corynebacterium* infection, as in the present case, steroid administration may facilitate relapse of GM through suppression of the host's immunity. It has also been demonstrated that patients with *Corynebacterium* infection tend to have longer treatment duration and higher risk of recurrence of complicated mastitis, compared with *Corynebacterium*-negative cases. Concurrent pregnancy and young age are both also associated with long treatment duration (13).

A recent meta-analysis comparing 138 cases of surgical treatment with 358 cases of steroid treatment demonstrated the superiority of the former (complete response rate: 90.6% vs. 71.8%; recurrence rate: 6.8% vs. 20.9%). Additionally, better results have been reported by combining surgery and steroid treatment, with a complete response rate of 94.5% and recurrence rate of 4.0% (14). Another meta-analysis showed that managing GM with steroids only may be less effective than a combination of steroids and surgery (15). Surgical procedures are associated with a different range of problems including stress and fear in patients, scarring and/or asymmetry of breast, and sometimes poorer cost-effectiveness. Furthermore, the main requirement for the surgical treatment option is that the lesion presents as a well-circumscribed mass. In diffuse lesions, it is not possible to excise the lesion while preserving the breast. In the present case, we selected surgical resection for the following reasons: (1) the patient was pregnant, and we were hesitant to use systemic steroid treatment; (2) bacterial culture was positive for *Corynebacterium* and steroid treatment would likely have been less effective; (3) the treatment period had already lasted for >2

months and the patient wanted early recovery; and (4) the lesion was in the axilla with a lesser impact on cosmesis.

In conclusion, there is no agreed standard management for GM. Therefore, treatment strategy should be planned on a case-by-case basis, taking into account the patient's situation. Unnecessary surgery should be avoided but early addition of surgical interventions may yield good outcomes, especially in pregnant women because of limited treatment options.

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