

Bilateral Xanthogranuloma of the Breast

Radiologic Findings and Pathologic Correlation

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Axanthogranuloma (XG) is a rare benign histiocytic disorder that occurs mostly in infants and young children.¹ Approximately 15% of XGs occur in adults and usually appear as solitary lesions in the head and the neck region. In most cases, an XG appears as a solitary yellow-red cutaneous nodule in the head and the neck region that regresses spontaneously over a course of months to years.^{1,2} An extracutaneous XG is uncommon, and it can occur with or without a cutaneous XG. An extracutaneous XG most commonly occurs in the eye, but findings at other sites such as the central nervous system, lungs, liver, spleen, kidneys, and bone have also been reported.²⁻⁵ The pathogenesis of XGs is unknown. Although physical and infectious factors have been considered, most investigators postulate that they are caused by a reactive response to an unknown stimulus.^{3,4} Xanthogranulomas can be divided into juvenile and adult forms. Distinctive from the juvenile form, the adult XG rarely undergoes a spontaneous regression, and concomitant extracutaneous examples are very uncommon.²⁻⁵ Patients with adult XGs are mostly in their late teens to late 30s, although approximately 5% of the patients are in their 60s to 70s.²⁻⁵ We report an exceptional case of an adult breast XG in a 60-year-old woman.

Abbreviations

BI-RADS, Breast Imaging Reporting and Data System;
XG, xanthogranuloma

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Case Report

A 60-year-old woman visited our institute for screening mammography. She had a history of diabetes mellitus for 3 years. On the screening mammography, approximately 3-cm irregularly shaped, high-density masses were found bilaterally. The masses were located in the subareolar area and the deep portion of the breasts. There were pleomorphic dense calcifications associated with the masses in the bilateral breasts (Figure 1A). Additional breast sonography revealed the irregular hypoechoic masses with a spiculated margin and marked posterior shadowing in the bilateral breasts (Figure 1B). The masses were located in the bilateral subareolar area. The radiologic findings were prospectively assessed according to the American College of Radiology Breast Imaging Reporting

and Data System (BI-RADS).⁶ These lesions were categorized as BI-RADS category 4b (intermediate suspicion for malignancy).

For investigating the patient's history, her old films were reviewed and compared with current examinations. Three years previously, she had undergone mammography and breast sonography. Breast sonography showed no remarkable change in the size, shape, margin, and echo texture of the lesions in both breasts (Figure 2A); however, mammography showed that the number and size of calcifications within the masses had increased compared with previous mammography (Figure 2B). Blood flow within the masses in both breasts was not shown (Figure 2C).

The patient had refused biopsy 3 years previously. We explained the suggestive finding of the lesions, and she consented to biopsy this time; hence, sonographically guided biopsy was per-

Figure 1. Screening mammography and breast sonography of a 60-year-old woman. A mediolateral oblique mammogram (A) and breast sonogram (B) of the bilateral breasts show irregular masses with the spiculated margin and clustered calcifications, which increased in size and number since the previous mammography.

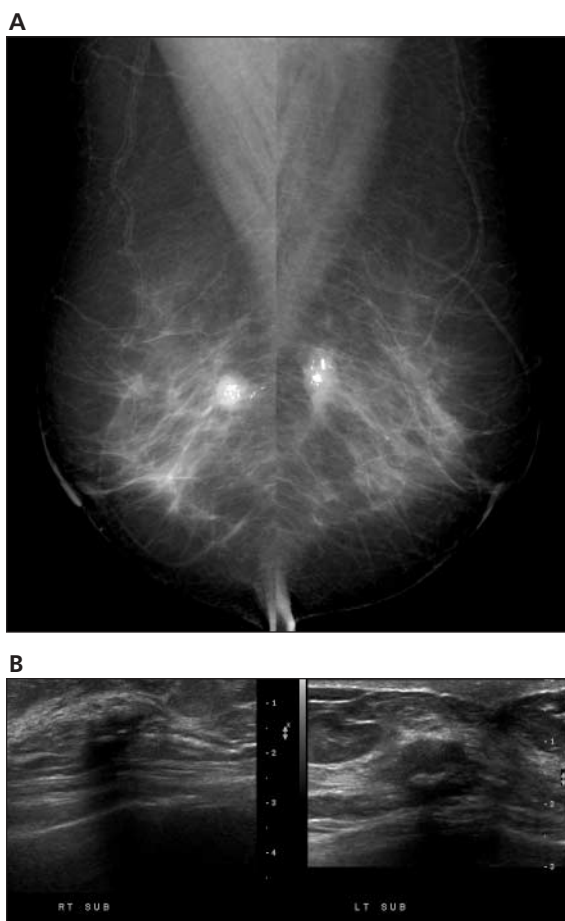
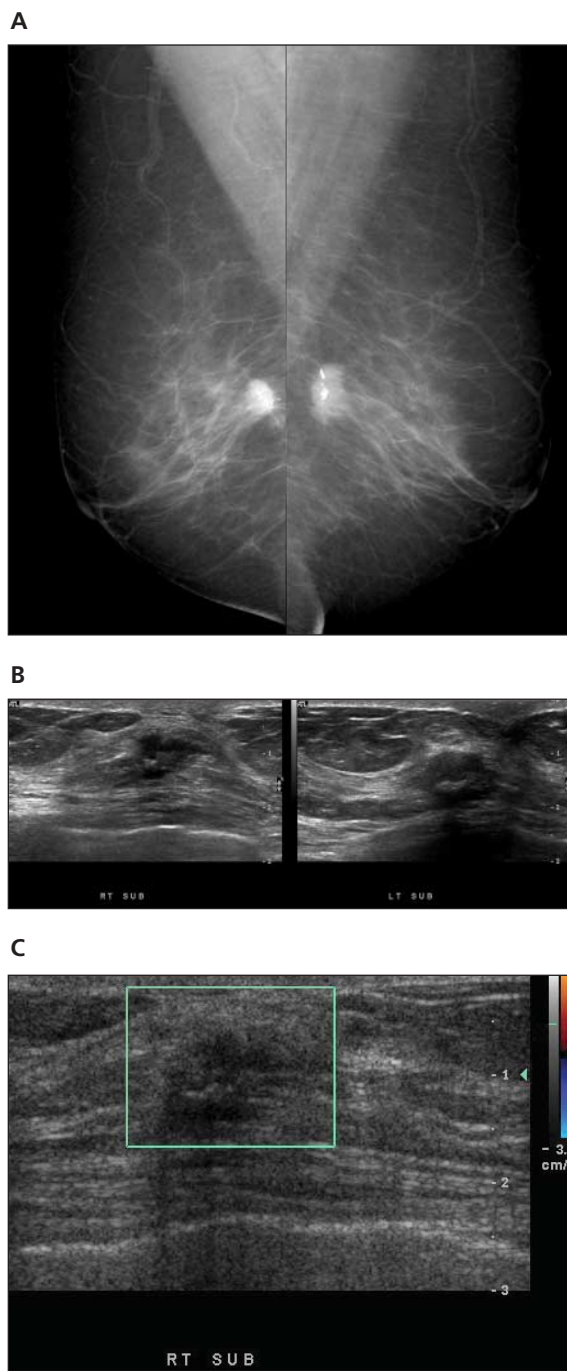


Figure 2. A previous mammogram (A) and breast sonogram (B) show irregular masses with the spiculated margin and clustered calcifications. Doppler sonography (C) shows no evidence of increased blood flow within a hypoechoic mass.



formed. An automated biopsy gun and a 14-gauge biopsy needle were used to obtain the cores of bilateral masses, and each biopsy was performed 5 times. The core biopsy specimens were pathologically examined and found to be composed of solid collections of histiocytes filled with a cholesterol-like lipid material. The biopsy results were consistent with an XG. Afterward, on the patient's request, these lesions were excised after wire needle localization. The excisional specimen had dense collagenous fibrosis, calcifications with conglomerated clusters of foamy histiocytes, and fibrosis with foreign body reaction. On the basis of these results, the diagnosis was confirmed as an XG (Figure 3).

Discussion

An adult-onset XG is a benign disorder showing spindle cells arranged in fascicles with minimal cellular pleomorphism admixed with inflammatory cells and histiocytes.¹ An XG is most likely to be confused with 2 entities, a metaplastic spindle cell carcinoma and an inflammatory pseudotumor. Examples of extracutaneous adult XGs are even more uncommon because most occur in the eye.⁴ To our knowledge, only 1 XG involving the breast has been reported.⁷ Few examples of extracutaneous adult XGs have had concomitant skin lesions and showed spontaneous regression.³⁻⁵ In our case, the patient had nonpalpable masses in both breasts without cutaneous

lesions, which may have existed for 3 years. In an adult XG occurring in the eye, computed tomography and magnetic resonance imaging showed infiltrating soft tissue masses without calcifications.⁸ The common radiologic findings of breast XGs have not been known. The appearance of the masses in our case was categorized as BI-RADS category 4b (intermediate suspicion for malignancy) on mammography and breast sonography. The increasing number of clustered amorphous calcifications had raised the suspicion for malignancy through the follow-up mammogram. However, it was difficult to distinguish a breast XG from a primary breast malignancy by only the radiologic findings. Hence, pathologic examinations by biopsy were necessary to confirm the breast XG.

In summary, we report a case of an adult XG involving the breast that showed no spontaneous regression. In the radiologic studies, including mammography and sonography, the finding of the breast XG was similar to that of a primary breast malignancy. Therefore, pathologic correlation was needed for the correct diagnosis.

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Figure 3. Microscopic findings show a diffuse nodular aggregation of histiocytes intermingled with chronic inflammatory cells (hematoxylin-eosin, original magnification $\times 100$).

