
CASE REPORT

Radiological and Positron Emission Tomography–Computed Tomography Features of Xanthogranulomatous Mastitis: An Extremely Rare Malignancy Mimicker in the Male Breast

T Wong¹, S Yang², WY Fung¹, RLS Chan¹, CM Chau¹, AWT Yung¹, JKF Ma¹

¹Department of Radiology, Princess Margaret Hospital, Hong Kong SAR, China

²Department of Radiology, Tuen Mun Hospital, Hong Kong SAR, China

CASE REPORT

A 71-year-old male presented with an incidental finding of a 1.6-cm hypermetabolic oval nodule in the subareolar region of the left breast with maximum standardised uptake value of 3.5 on positron emission tomography–computed tomography (PET-CT) [Figure 1]. He had stage IV colon cancer with multiple liver metastases. No abnormal lymph node was seen in the left axilla. Physical examination revealed a 1.5-cm firm mobile mass at the left breast subareolar region that was considered clinically suspicious.

Mammography and tomosynthesis images revealed an oval circumscribed lesion of equal-to-high density at the subareolar region of the left breast with no nipple or skin retraction (Figure 2). There was no associated suspicious calcification or architectural distortion. On ultrasound, a 1.8 cm × 0.8 cm × 1.6 cm oval circumscribed complex cystic and solid lesion with posterior acoustic enhancement was seen at the subareolar region of the

left breast (Figure 3a). A 0.3-cm echogenic mural nodule was seen at the anterior aspect of the lesion (Figure 3b). Vessels in the rim were noted on colour Doppler (Figure 3c). No abnormal axillary lymphadenopathy was found. The lesion was classified as BI-RADS (Breast Imaging Reporting and Data System) category 4B.

Core biopsy revealed xanthogranulomatous mastitis (XGM) without malignancy. The patient opted for conservative management. He remained asymptomatic for the breast lesion upon follow-up 6 months later, with only a vague lump in the subareolar region of the left breast palpable during physical examination. Ultrasound revealed interval shrinkage of the lesion to 1.3 cm × 0.7 cm × 1.1 cm. It remained oval and circumscribed, with mixed cystic and solid components (Figure 4).

DISCUSSION

XGM is a rare chronic benign inflammatory condition of the breast, with fewer than 30 cases reported and

Correspondence: Dr T Wong, Department of Radiology, Princess Margaret Hospital, Hong Kong SAR, China
Email: gloria_wong@live.com

Submitted: 21 Dec 2021; Accepted: 26 Jan 2022.

Contributors: All authors designed the study. TW, SY, WYF, RLSC and CMC acquired and analysed the data. TW and SY drafted the manuscript. All authors critically revised the manuscript for important intellectual content. All authors had full access to the data, contributed to the study, approved the final version for publication, and take responsibility for its accuracy and integrity.

Conflicts of Interest: All authors have disclosed no conflicts of interest.

Funding/Support: This study received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Data Availability: All data generated or analysed during the present study are available from the corresponding author on reasonable request.

Ethics Approval: This study was approved by the Kowloon West Cluster Research Ethics Committee of Hospital Authority, Hong Kong (Ref No.: 152-12). The patient was treated in accordance with the tenets of the Declaration of Helsinki and provided written informed consent for the investigations and the publication of this case report.

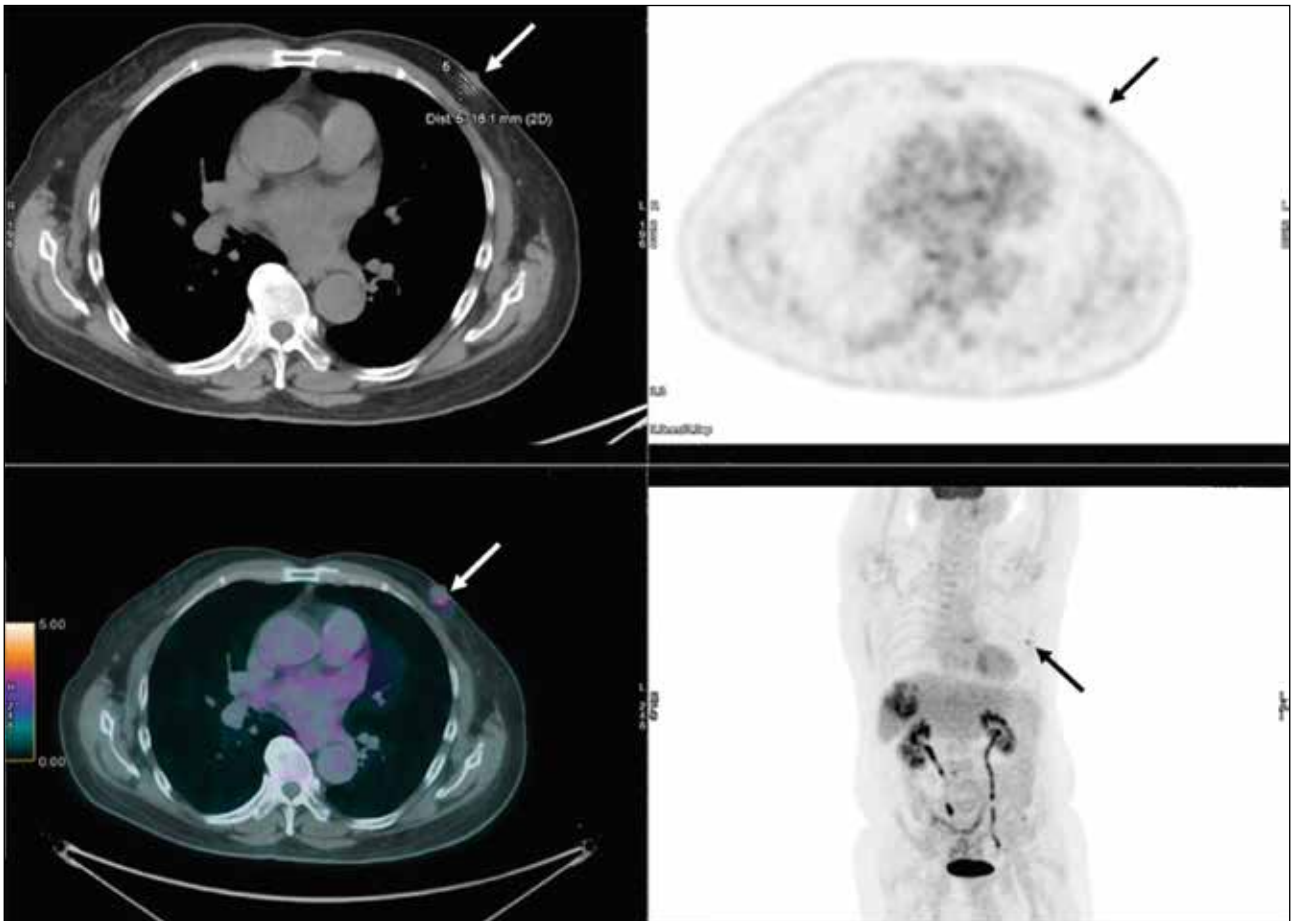


Figure 1. Positron emission tomography–computed tomography of the patient for colon cancer workup incidentally revealed a 1.6-cm hypermetabolic lesion at the subareolar region of the left breast (arrows).

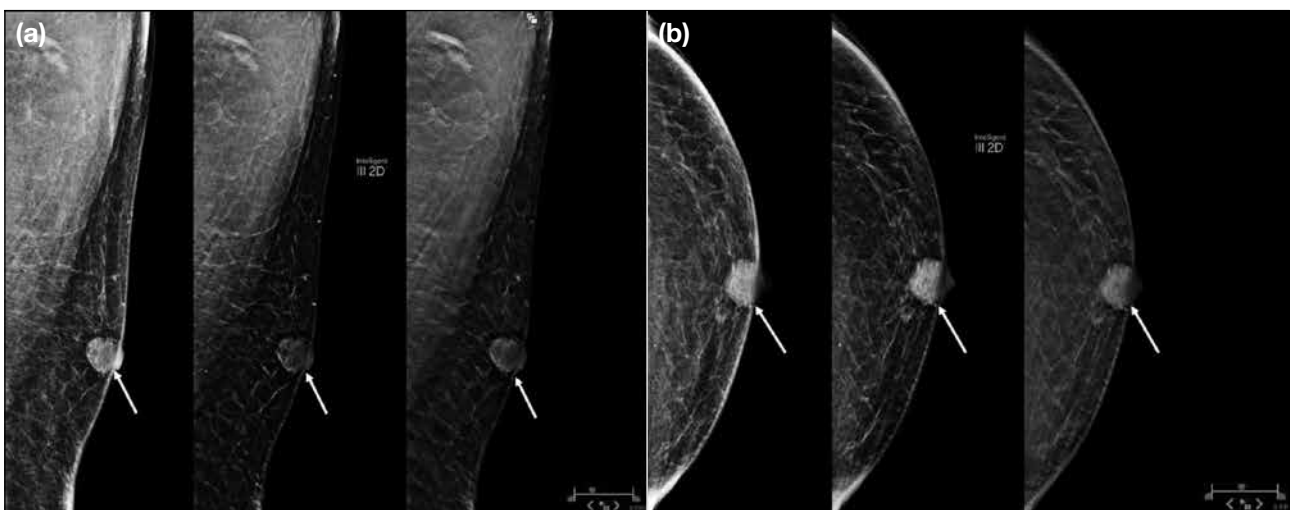


Figure 2. Full-field digital mammogram, synthetic two-dimensional mammogram, and tomosynthesis image of (a) mediolateral oblique and (b) craniocaudal views demonstrated an oval circumscribed equal-to-high density lesion at left breast subareolar region (arrows).

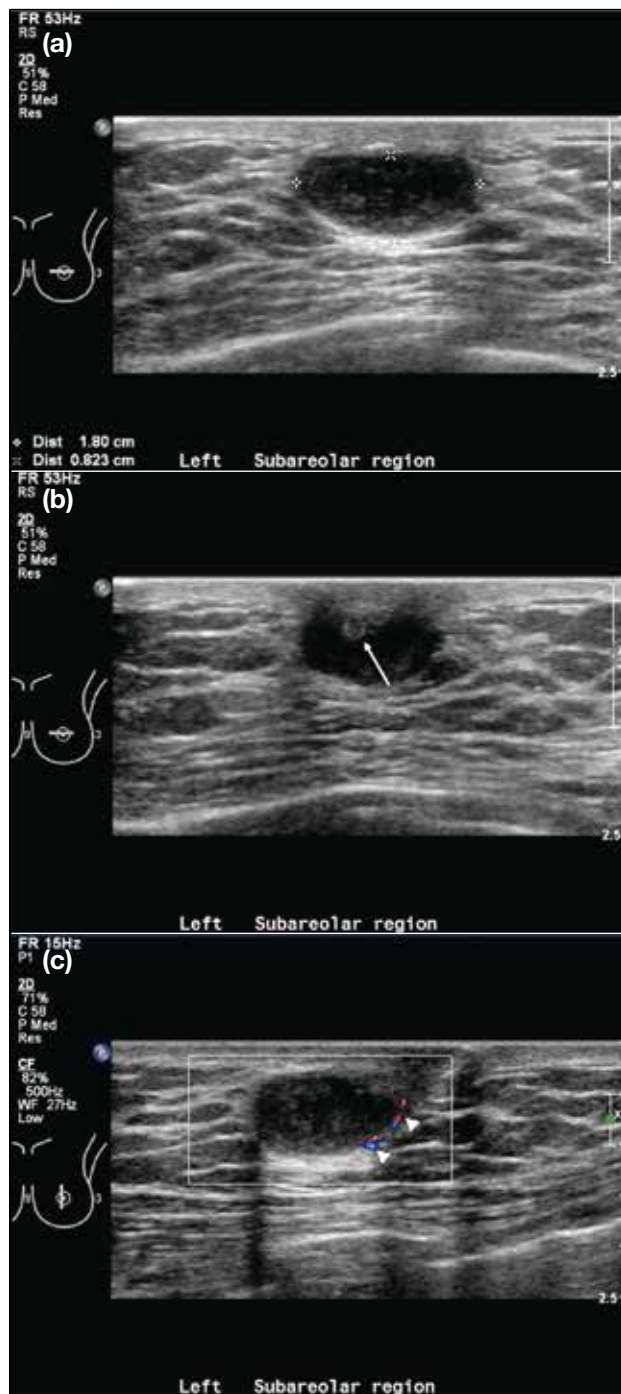


Figure 3. (a) Ultrasound of left breast showed a 1.8-cm oval circumscribed complex cystic and solid lesion with posterior acoustic enhancement (marked with crosses). (b) A mural nodule was seen over its anterior aspect (arrow). (c) Vessels in the rim (arrowheads) were noted on colour Doppler.

none in men since it was first described in 2005.^{1,2} To the best of our knowledge, this is the first reported case in a male XGM patient with PET-CT and digital breast tomosynthesis performed.



Figure 4. Follow-up ultrasound of left breast 6 months later showed interval shrinkage of the lesion to 1.3 cm in size. (a) It remained oval and circumscribed with cystic and solid components (marked with crosses). (b) The echogenic mural nodule (arrow) was seen.

The aetiology of XGM remains debatable. Koo and Jung³ postulated it to be a result of obstruction and rupture of dilated ducts with foamy histiocyte aggregates. XGM has been reported in patients with concurrent breast cancer,³ fat necrosis,³ lactational changes,³ and implant rupture,^{4,6} suggesting that prior insult, hypersecretory status or implant leak might be triggering factors.^{3,5} It has also been reported in breast cancer regression following neoadjuvant chemotherapy and in Erdheim–Chester disease with breast involvement.^{7,8}

XGM is commonly identified as a BI-RADS category 4 or 5 lesion on breast imaging, as noted in the present case.^{1,3,9,10} Nonetheless the common imaging features of XGM have not been well reported¹⁰ and were only described in a few cases. XGM lesions have been depicted as solid hypoechoic lesions on ultrasound,^{1,4,9,10}

with most described as irregular.^{1,9,10} On mammography, XGM has been revealed as a poorly defined dense area⁹ or as a spiculated mass with increasing number and size of the associated clustered calcifications.¹⁰ Previous reports of magnetic resonance imaging described a patient with a poorly defined heterogeneous area in the breast that showed early and heterogeneous enhancement with upward curve,⁹ and another patient with prior breast reconstruction presenting with areas of contrast enhancement in the breast reconstruction and latissimus dorsi scar, mimicking breast cancer recurrence.⁵

In our patient, XGM presented initially as a hypermetabolic lesion on PET-CT. It was an oval circumscribed lesion of equal-to-high density on full-field digital mammography and digital breast tomosynthesis and was complex mixed cystic and solid with an echogenic mural nodule and vessels in the rim on ultrasound. Such radiological features have not been reported before. Nonetheless XGM has previously been reported to have a cystic precursor stage, presumed to represent the markedly dilated duct preceding its rupture.¹ It is evident that XGM can have a variable radiological presentation.

A complex cystic breast lesion can have a wide range of differential diagnoses, but there is a substantial risk that it is a malignant lesion, such as ductal carcinoma in situ or invasive ductal carcinoma and invasive lobular carcinoma.¹¹ Other common diagnoses include fibrocystic changes, fibroadenoma, intraductal or intracystic papilloma with or without atypia, atypical ductal hyperplasia, and lobular neoplasia.¹¹ It is therefore radiologically difficult to differentiate this benign entity from a malignant condition. It has been suggested that the mass-forming nature of XGM with the presence of inflammation accounts for its suspicious appearance,⁹ therefore biopsy is often required to confirm the diagnosis.^{1,10,11}

XGM is often asymptomatic with a self-limiting course.¹ It was an incidental finding in our case, with subsequent

shrinkage. Nonetheless there is insufficient evidence to guide optimum management and radiological follow-up.¹ Surgical excision is not common practice but may be required for large or infiltrative lesions since drainage, steroids, and antibiotics are unhelpful.¹

In conclusion, XGM is exceedingly rare, and our patient is the first reported case in a male. Its radiological appearance can mimic malignancy and it may demonstrate hypermetabolism on PET-CT. Histological evaluation is essential to confirm the diagnosis of this benign entity.

REFERENCES

1. Kapoor H, Zhang Y, Qasem SA, Owen W, Szabunio MM. Xanthogranulomatous mastitis preceded by cysts on ultrasound: two cases with review of literature. *Clin Imaging*. 2021;78:64-8.
2. Shin SJ, Scamman W, Gopalan A, Rosen PP. Mammary presentation of adult-type "juvenile" xanthogranuloma. *Am J Surg Pathol*. 2005;29:827-31.
3. Koo JS, Jung W. Xanthogranulomatous mastitis: clinicopathology and pathological implications. *Pathol Int*. 2009;59:234-40.
4. Dinets A, Unukovych D, Khrapach V, Zakhartseva O, Sulik V, Kaminskyi E, et al. An unusual case of a ruptured Poly Implant Prothèse breast implant associated with xanthoma. *Case Reports Plast Surg Hand Surg*. 2016;3:11-5.
5. Hussain T, Elahi B, Long E, Mahapatra T, McManus PL, Kneeshaw PJ. Xanthogranulomatous inflammation involving latissimus dorsi donor site and implant breast reconstruction: case report and literature review. *World J Surg Oncol*. 2012;10:166.
6. Hernanz F, Baeza S, Serna E, Gómez-Fleitas M. Xanthogranulomatous capsulitis mimicking a polypoid neoplasm disease: an unusual presentation of ruptured Poly Implant Prothèse (PIP) breast implant. *Eur J Plas Surg*. 2013;36:797-800.
7. Aktepe F, Kapucuoğlu N, Pak I. The effects of chemotherapy on breast cancer tissue in locally advanced breast cancer. *Histopathology*. 1996;29:63-7.
8. Barnes PJ, Foyle A, Haché KA, Langley RG, Burrell S, Juskevicius R. Erdheim-Chester disease of the breast: a case report and review of the literature. *Breast J*. 2005;11:462-7.
9. de Oliveira I Junior, Viana CR, Sabino SM, Kerr LM, Vieira RA. Xanthogranulomatous mastitis mimicking locally advanced breast cancer. *Breast J*. 2017;23:227-9.
10. Hwang SH, Son EJ, Oh KK, Kim EK, Jung J, Jung WH. Bilateral xanthogranuloma of the breast: radiologic findings and pathologic correlation. *J Ultrasound Med*. 2007;26:535-7.
11. Doshi DJ, March DE, Crisi GM, Coughlin BF. Complex cystic breast masses: diagnostic approach and imaging-pathologic correlation. *Radiographics*. 2007;27 Suppl 1:S53-64.