Case Report | Surgery: Breast Surgery

Granular cell tumor of the male breast: a case report and review of the literature

Olivia W. Galloway, Ned Z. Carp

Lankenau Medical Center, Wynnewood, PA, USA

Contributions: (I) Conception and design: Both authors; (II) Administrative support: NZ Carp; (III) Provision of study materials or patients: NZ Carp; (IV) Collection and assembly of data: Both authors; (V) Data analysis and interpretation: Both authors; (VI) Manuscript writing: Both authors; (VII) Final approval of manuscript: Both authors.

Correspondence to: Olivia W. Galloway, MD. Lankenau Medical Center, 100 E. Lancaster Ave, MOB East Suite 422, Wynnewood, PA 19083, USA. Email: olivia.w.galloway@gmail.com.

Background: Granular cell tumors (GCTs) of the breast are rare tumors that originate from Schwann cells. They are most common in premenopausal females of African descent, and fewer than 30 cases of GCT in males have been described in the literature. Most prior cases of male breast GCTs presented as symptomatic palpable masses in the breast that are subsequently excised. In this case, we describe a rare asymptomatic presentation of male GCT that was completely excised on core needle biopsy and found to be associated with a granuloma after excisional biopsy, the first case reported in the literature.

Case Description: A 56-year-old African American male with a history of hypertension, alcohol abuse, atrial fibrillation, heart failure, diabetes, and pancreatitis incidentally found to have a mass in his right breast while undergoing diagnostic imaging workup for a mass in his contralateral breast. Biopsy revealed a GCT, positive for S-100 and CD68. He underwent excisional biopsy of his GCT, the lesion was completely excised, and he is recovering well with no evidence of recurrence at 6 months.

Conclusions: GCTs of the male breast are rare entities that present diagnostic and therapeutic challenges for providers and mimic breast carcinomas. The pathophysiology of GCTs is unknown, and several risk factors are postulated to play a role in development of GCTs, such as gynecomastia and alcohol abuse in this patient. This case provides further insight into the factors leading to development of male breast GCTs, and details appropriate diagnosis and management of this disease.

Keywords: Granular cell tumor (GCT); male breast; case report

Received: 06 September 2023; Accepted: 18 March 2024; Published online: 17 May 2024.
doi: 10.21037/amj-23-171
View this article at: https://dx.doi.org/10.21037/amj-23-171

Introduction

Background

Granular cell tumors (GCTs) are rare neoplasms of Schwann cell origin that may arise in any anatomic location, most commonly the skin and subcutaneous soft tissues (1). GCTs of the breast are rare; approximately 5–15% of all GCTs are in the breast and 0.1–0.7% of all breast tumors are GCTs (2). Male patients account for 6.6% of breast GCTs (2).

Rationale and knowledge gap

The histogenesis of GCTs is unknown and presents a diagnostic and therapeutic challenge due to its rarity and similarity on imaging to other breast neoplasms. Prior reported cases of male GCTs presented as symptomatic palpable breast masses. Here, we present a case of a man incidentally diagnosed with breast GCT while undergoing workup for a breast mass on the contralateral side. After breast GCT was diagnosed on core needle biopsy, he underwent excisional biopsy, which showed complete

excision of the GCT from core needle biopsy and an associated granuloma. The complete excision from core needle biopsy as well as the association of breast GCT and granuloma has not been described in the literature previously. After one year of follow-up, he has no evidence of recurrence on physical examination, and will continue to follow-up annually.

Objective

This case describes a rare GCT in a male breast, and adds to previously existing knowledge regarding risk factors for development of male breast GCTs, presentation, and treatment options. Additionally, the association with granuloma has not been previously described, and clinical significance remains unknown. We present this article in accordance with the CARE reporting checklist (available at https://amj.amegroups.com/article/view/10.21037/amj-23-171/rc).

Highlight box

Key findings

- Most cases of male breast granular cell tumors (GCTs) present as palpable masses of the breast.
- This case describes a rare asymptomatic case of male breast GCT that was incidentally discovered and completely excised using core needle biopsy.
- This case highlights potential risk factors such as alcohol use and angiotensin converting enzyme (ACE) inhibitor usage that may be implicated in the development of male GCTs.

What is known and what is new?

- There are approximately 30 cases of male GCTs reported in the literature, and it is difficult to ascertain what factors may lead to the development of this disease.
- It has been suggested that medication usage or lifestyle factors increasing the risk of gynecomastia may contribute to the development of male breast GCTs. This case highlights a patient with multiple risk factors for gynecomastia who had developed an asymptomatic breast GCT and granuloma.
- In this case, the GCT was found to be completely excised during core needle biopsy and associated with a granuloma, which has not been previously reported.

What is the implication, and what should change now?

- GCTs may be mimickers of breast carcinoma on physical exam and imaging.
- Understanding risk factors for GCTs will enable providers to accurately diagnose and treat GCTs.
- Small incidentally found GCTs may be completely excised with core needle biopsy, however, recurrence rates are unknown.

Case presentation

The patient is a 56-year-old African American male with past medical history of hypertension on an angiotensin converting enzyme (ACE) inhibitor, alcohol abuse on folic acid and thiamine, atrial fibrillation on Eliquis, heart failure with reduced ejection fraction, poorly controlled insulin-dependent diabetes, and pancreatitis who presented to his primary care physician on 15 July 2022 with a palpable mass of his left breast and breast pain. He did not have any palpable masses in his right breast. He had no personal or family history of breast cancer. He underwent a bilateral diagnostic mammogram on 1 August 2022 which showed a small focal mass at 9:00–9:30 in the right breast mid-depth and global asymmetry on the left consistent with benign gynecomastia (*Figure 1*). Bilateral breast ultrasound showed a 0.5×0.4×0.5 cm mass in the right breast at 9:00, 1 cm from the nipple with slightly indistinct margins and mild posterior acoustic shadowing that corresponded to the mass seen on mammogram (*Figure 1*). Due to the findings on the right, the final assessment was Breast Imaging Reporting and Data System (BIRADS)-4, suspicious abnormality, for which a biopsy was recommended. He was referred to the nurse navigator for surgical consultation and a biopsy. There were no financial or cultural diagnostic challenges.

The patient was then referred to surgical oncology and saw the surgical oncologist in the office on 11 August 2022. An ultrasound-guided fine needle aspiration and core biopsy of the lesion was performed. Core needle biopsy revealed a GCT (*Figure 2*). The lesional cells were positive for S-100 and CD68, and negative for CD34, desmin, pancytokeratin, estrogen receptor, and progesterone receptor. He then underwent a right breast ultrasound-guided wire localized excisional biopsy of his GCT on 16 September 2022. There were no changes to the planned surgery. The specimen contained a 0.6×0.5×0.4 cm white firm irregular markedly ill-defined possible mass located 0.4 cm from the medial margin, 0.5 cm from the lateral margin, 1.2 cm from the posterior margin, 1.9 cm from the anterior margin, and 1.3 cm from the inferior margin. Anterior to the mass was a 0.5×0.5×0.4 cm well-circumscribed ovoid cystic cavity grossly consistent with a biopsy cavity containing the coiled biopsy clip. Final pathology revealed a palisading granuloma with adjacent histiocytic deposits consistent with the previous biopsy. There was no evidence of malignancy, including at the resection margin. On immunohistochemical staining, CD68 was positive, while S-100 and estrogen receptor were negative. The patient recovered from

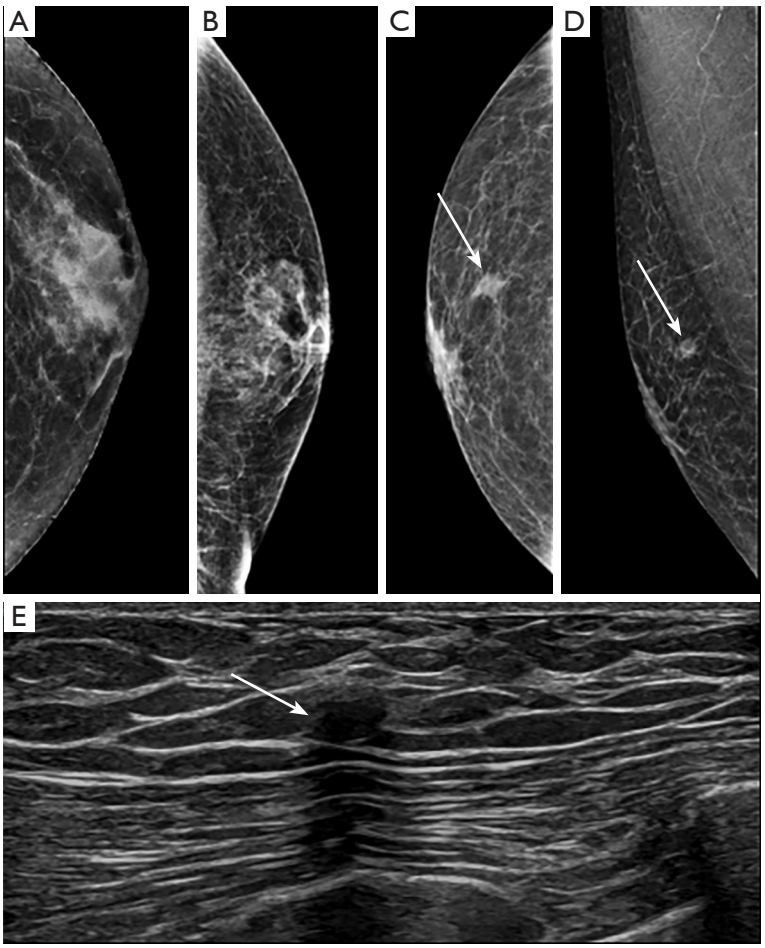

Figure 1 Mammographic and ultrasound images of breast mass. (A) Left breast craniocaudal image showing scattered areas of fibroglandular density without discrete mass. (B) Left breast mediolateral oblique view. (C) Right breast craniocaudal image, arrow pointing to small focal mass. (D) Right breast mediolateral oblique image, arrow pointing to small focal mass. (E) Ultrasonographic images of hypoechoic round right breast mass (arrow) with posterior acoustic shadowing measuring 0.5×0.4×0.5 cm corresponding to mammogram at 9:00, 1 cm from the nipple with slightly indistinct margins.

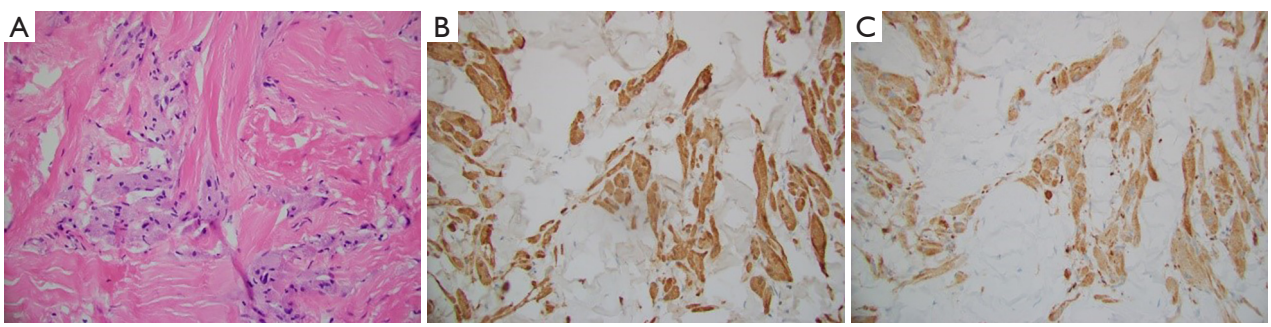

Figure 2 Microscopic views of the granular cell tumor after core needle biopsy. (A) Hematoxylin-eosin stain showing sheets of polygonal cells with round to oval eccentric nuclei, ×40 magnification. (B) GCT cells staining positive for S-100, ×40 magnification. (C) GCT cells staining positive for CD-68, ×40 magnification. GCT, granular cell tumor.

his operation and was doing well on his postoperative examination on 30 September 2022. His 8-month follow-up visit on 2 May 2023 showed no evidence of recurrence on clinical exam. He will follow-up with surgical oncology in one year. There were no adverse or unanticipated events during his care.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

First described by Abrikossoff in 1926, GCTs are rare tumors arising from Schwann cells arising in varied anatomic locations (3). They occur more commonly in premenopausal women and in African American patients (4). GCTs of the breast in men are rare; there have been approximately thirty case reports of GCTs in males in the literature (summarized in *Table 1*) (4-10). This was a case of an African American male who had an incidentally found asymptomatic benign GCT that was associated with a granuloma.

GCTs of the breast have a variable presentation; usually a painless, solid, firm, poorly circumscribed mass (4). They may cause skin retraction or adhesion to the pectoralis due to their infiltrating growth pattern, leading clinicians to be concerned for breast cancer (4). In prior cases, most GCTs in the male breast were palpable masses measuring 1 to 3 cm (*Table 1*). However, in this case, the GCT was incidentally found during workup for a palpable mass on the contralateral breast. This led to diagnosis of a smaller GCT than previously reported, measuring 0.5 cm and was subsequently found to be completely excised during core needle biopsy.

During workup for GCTs, imaging patterns on mammography or ultrasound may also be variable. Imaging findings may include heterogenous echo texture, indistinct or spiculated margins, increased blood flow, and posterior acoustic shadowing, which may mimic breast carcinoma (8). In our case, a mass with slightly indistinct margins and mild acoustic shadowing was seen, which was indeterminate for malignancy. Although GCTs may have features that mimic carcinoma on imaging, tissue diagnosis is required to differentiate between these two entities.

On pathologic examination, GCT cells show granular cytoplasm and strong cytoplasmic and nuclear staining for S-100 (4). They also stain positive for neuron specific enolase, CD68, and CKI-C3. They typically stain negative for estrogen receptor and progesterone receptor (4). In this case, the GCT stained positive for S-100 and CD68 on core needle biopsy. After the GCT was excised, there was no residual GCT in the specimen, as demonstrated by negative staining for S-100, indicating complete excision from the core needle biopsy. The mass seen on ultrasound and mammogram likely correlated with the granuloma, not the GCT, indicating that the GCT was radiographically occult. Additionally, this GCT was found to be associated with a granuloma upon final pathology review after excisional biopsy. Granulomas in male breasts are also rare entities and may occur as a result of cancer, sutures, sarcoidosis, tuberculosis, or cholesterol (11-13). The cause and clinical significance of the granuloma in this patient is unclear, though it may have been a response to the adjacent GCT. There have been several case reports of male idiopathic granulomatous mastitis in association with gynecomastia (14). Our patient was diagnosed with gynecomastia which may have been a risk factor for the development of his GCT and granuloma. He had multiple risk factors for development of gynecomastia, such as alcohol abuse and ACE inhibitor usage, which may place him at higher risk for development of benign and malignant breast conditions.

Treatment of benign GCT of the breast consists of surgical excision. Benign GCTs have a recurrence rate of 2–8% when resection margins are free of tumor and 20% if margins are positive for tumor (15). Annual follow-up is advised. In less than 1% of cases, breast GCTs are malignant. Malignant GCTs are aggressive, and recurrence rates are as high as 32% with a 39% 3-year mortality rate (15). Metastases are also common in malignant GCTs (50%), and most common sites are lung, liver, bone, and axillary lymph nodes (15). It is important to consider the malignant and aggressive potential when encountering GCTs in men, and to ensure patients are followed closely. At postoperative visit and 6-month follow-up the patient reported relief that his GCT was excised and understood the necessity to continue to undergo additional annual surveillance.

The identification of several potential risk factors in the development of rare breast pathologies in males is a strength of this study. One weakness of this study is the short period of follow-up for this patient, less than a year since diagnosis. The GCT was found to be completely excised during his core needle biopsy, and it is unclear what the margins from

Table 1 Summary of cases of male breast GCT seen in the literature to date

Number	First author	Year	Age (years)	Clinical impression	Size (cm)	Surgical treatment
1	Simon	1947	60	Benign tumor suspected	2.5	Local excision
2	Peison	1964	21	Abscess suspected	1.8	Local excision
3	Mulcare	1968	35	Carcinoma	2	Local excision
4	Umansky	1968	44	Carcinoma	2.5	Local excision
5	Umansky	1968	55	Carcinoma	NA	Radial mastectomy
6	Hart	1973	36	Carcinoma	3	Local excision
7	Sussman	1973	56	Carcinoma	1.8	Local excision
8	Zemoura	1984	32	Malignant tumor	2.2	Radial mastectomy
9	Zemoura	1984	49	Malignant tumor	NA	Local excision
10	Demay	1984	26	NA	1.5	NA
11	Townsend	1985	29	Fibroadenoma	1.5	Local excision
12	Khansur	1987	NA	NA	NA	Local excision
13	Baeten	1992	49	Malignancy suspected	2	Local excision
14	Damiani	1993	25	Malignancy suspected	2	Local excision
15	Takahashi	1995	49	NA	2	Local excision
16	Reale	1995	38	Carcinoma	2.7	Local excision
17	Mariscal	1995	47	Carcinoma	1.7	Local excision
18	Placidi	1995	30	NA	1.5	Local excision
19	Rogall	1995	35	Carcinoma suspected	1	Local excision
20	Okuda	1996	45	Carcinoma	2	Local excision
21	Lee	1997	35	Carcinoma	3	Local excision with lymphadenectomy
22	Adeniran	2004	32	NA	0.8	NA
23	Adeniran	2004	36	NA	1	NA
24	Adeniran	2004	42	NA	2.5	NA
25	Kim	2011	49	Carcinoma suspected	1.7	Local excision
26	Kim	2011	50	Carcinoma suspected	1.7	Local excision
27	Taglietti	2011	54	Carcinoma suspected	2.5	Local excision
28	Patel	2013	43	Carcinoma suspected	2.2	Local excision
29	Kuo	2019	NA	Carcinoma suspected	1.7	NA
30	Abreu	2020	57	–	1.2	NA

GCT, granular cell tumor; NA, not available.

the GCT were. His GCT was likely radiographically occult, but completely excised. He will need longer term follow-up to determine if he will have a recurrence. It is also not clear what the clinical significance or etiology of his granuloma is, and more cases and studies will have to be performed to determine if there is any association between granulomas and GCTs. It is also likely that a subset of the population is also harboring radiographically occult GCTs that are not excised, and potentially not require excision, despite a small chance of malignancy.

Conclusions

This case contributes to the limited existing literature regarding male breast GCTs and male breast granulomas. It highlights a rare case of an asymptomatic GCT in association with a granuloma in a patient with gynecomastia due to multiple factors such as alcohol abuse and ACE inhibitor usage. Additionally, upon pathology review after his excisional biopsy, his GCT was found to be completely excised, likely during his core needle biopsy. The patient will require long term follow-up to determine if he has any recurrence. He additionally had a granuloma that was found on excisional biopsy for his GCT, the etiology and clinical implications of which is unknown.

Acknowledgments

Funding: None.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://amj.amegroups.com/article/view/10.21037/amj-23-171/rc

Peer Review File: Available at https://amj.amegroups.com/article/view/10.21037/amj-23-171/prf

Conflicts of Interest: Both authors have completed the ICMJE uniform disclosure form (available at https://amj.amegroups.com/article/view/10.21037/amj-23-171/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

References

1. Mobarki M, Dumollard JM, Dal Col P, et al. Granular cell tumor a study of 42 cases and systemic review of the literature. Pathol Res Pract 2020;216:152865.
2. Brown AC, Audisio RA, Regitnig P. Granular cell tumour of the breast. Surg Oncol 2011;20:97-105.
3. Abrikossoff A. Über Myome: ausgehend von der quergestreiften willkürlichen Muskulatur. Virchows Arch path Anat 1926;260:215-33.
4. Adeniran A, Al-Ahmadie H, Mahoney MC, et al. Granular cell tumor of the breast: a series of 17 cases and review of the literature. Breast J 2004;10:528-31.
5. Taglietti L, Vettoretto N, Blanzuoli L, et al. Granular cell tumor of the male breast. Updates Surg 2011;63:139-42.
6. Lee S, Morimoto K, Kaseno S, et al. Granular cell tumor of the male breast: report of a case. Surg Today 2000;30:658-62.
7. Kim EY, Kang DK, Kim TH, et al. Granular cell tumor of the male breast: two case descriptions and brief review of the literature. J Ultrasound Med 2011;30:1295-301.
8. Kuo F, Lally K, Lewis M, et al. Granular cell tumour in male breast mimicking breast carcinoma. BMJ Case Rep 2019;12:e227805.
9. Patel HB, Leibman AJ. Granular cell tumor in a male breast: mammographic, sonographic, and pathologic features. J Clin Ultrasound 2013;41:119-21.
10. Abreu N, Filipe J, André S, et al. Granular cell tumor of the breast: correlations between imaging and pathology findings. Radiol Bras 2020;53:105-11.
11. Siddiqui B, Habib Faridi S, Maheshwari V, et al. Granulomatous Response with Breast Cancer: A Case Report. Iran J Pathol 2016;11:171-5.
12. Nam G, Singer TM, Lourenco AP, et al. Cholesteroloma of the breast: A 10 year retrospective review of 79 cases with radiology correlation. Breast J 2019;25:1177-81.
13. Grove J, Meier C, Youssef B, et al. A Rare Case of Sarcoidosis Involving Male Breast Tissue. Cureus 2022;14:e21387.
14. Han YK, Ting W, Ting P, et al. Case report on gynecomastia with ipsilateral recurrence of

granulomatous lobular mastitis. J Int Med Res 2023;51:3000605231187815.
15. Swamy N, Jennings P, Taylor R, et al. Revisiting the Inscrutable Granular Cell Tumors in the Breast and Beyond: An Institutional Experience: The inscrutable granular cell tumor. Arch Breast Cancer 2022;9:204-12.

doi: 10.21037/amj-23-171
Cite this article as: Galloway OW, Carp NZ. Granular cell tumor of the male breast: a case report and review of the literature. AME Med J 2025;10:5.