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CASE REPORT

A Rare Complication of Silicone Breast Implant Rupture

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Silicone, Hypercalcemia, Lymph nodes, Steroids, Calcitriol, Granulomas, Thorax, Surgery, Parathyroid hormone, Hospital medicine

Abstract
Rupture of silicone breast implants is common with the time spent in situ but often goes undetected. We encountered a 55-year-old woman who presented with symptoms attributable to hypercalcemia after undergoing the removal of ruptured silicone breast implants. Investigations uncovered multiple enlarged intrathoracic lymph nodes accompanied by extensive granulomatous disease. Although systemic steroids helped to regulate her symptomatic hypercalcemia, it resurfaced on cessation of therapy. This patient's case underscores the necessity of monitoring silicone breast implants and being vigilant for complications that may arise from their rupture.

Background
Every year, millions of people worldwide, including approximately 275 000 in the United States (1), receive silicone breast implants. The US Food and Drug Administration has identified approximately 25 complications associated with silicone breast implants (2), with implant rupture being a significant concern due to its potential for systemic deleterious effects. Although the incidence of silicone breast implant rupture is unclear, the risk for rupture increases with the age of the implant.

Objective
We present this patient's case to emphasize the importance of monitoring silicone breast implants and staying alert for potential complications that can occur from their rupture.

Case Report
A 55-year-old woman presented to our clinic with a 6-week history of worsening upper back pain, fatigue, anxiety, dry mouth, abdominal bloating, constipation, and unintentional weight loss of more than 20 lbs. She had been taking calcitriol 1 mcg daily for 30 years for hypoparathyroidism, which developed after thyroidectomy for follicular thyroid carcinoma. Six weeks earlier, she had undergone extraction of ruptured bilateral silicone breast implants, which had been inserted approximately 35 years ago. During surgery, part of the fibrous capsule on the right side was left over in situ, as it was deeply adherent to the underlying musculature.

Before surgery, the patient had not undergone any routine follow-up breast imaging since the augmentation. However, a computed tomography scan of the chest was requested 8 weeks previously, when she recovered from COVID-19 illness and experienced atypical chest pain. This revealed the presence of curvilinear lines with an intact fibrous capsule on both sides, depicting collapsed implant shells floating in the silicone gel, the Linguine sign (3), of intracapsular breast implant rupture (Figure 1). The lung parenchyma and mediastinum were found to be normal on computed tomography scan. Her blood counts, liver and renal function tests, and electrolyte levels were all within the normal range in her previous tests before and after COVID-19, as well as before the breast surgery.

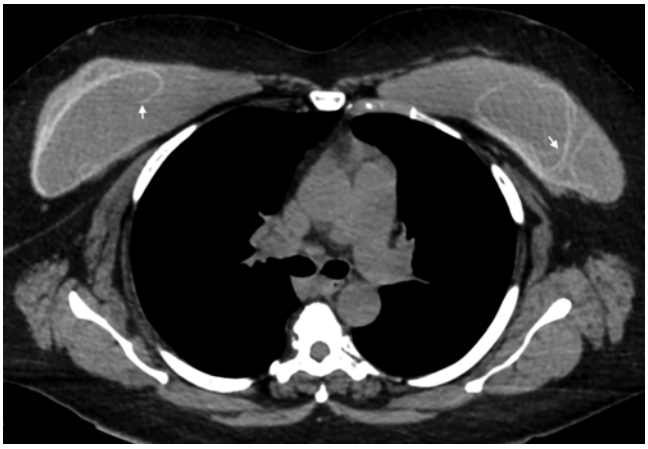

Figure 1. Computed tomography scan of chest, axial view, shows curvilinear lines in both breasts (*arrows*, Linguine sign) indicating intracapsular rupture of breast implants.

Findings of the physical examination were normal, but blood tests revealed high corrected serum calcium of 3.2 mmol/L (reference range 2.1–2.5), low parathyroid hormone level of 4 ng/L (reference range 4.6–58.0), normal phosphorus of 1.1 mmol/L (reference range 0.9–1.4), elevated creatinine of 194.5 μmol/L (reference range 65.4–119.3), normal bicarbonate of 25 mmol/L (reference range 22–26), and normal arterial blood pH of 7.39 (reference range 7.35–7.45). Her erythrocyte sedimentation rate was 28 mm/h (reference value up to 20 mm/h), and her serum angiotensin-converting enzyme level was 36 U/L (reference range 9–67 U/L). Magnetic resonance imaging scan of the chest revealed multiple enlarged mediastinal and hilar lymph nodes (Figure 2).

Despite the initial treatment with liberal intravenous and oral hydration and discontinuation of calcitriol, her symptoms, hypercalcemia, and renal function did not improve significantly. A mediastinoscopic lymph node biopsy revealed numerous noncaseating granulomas (Figure 3), and further examinations excluded malignancy, mycobacteria, and fungi.

Symptomatic hypercalcemia and resultant acute kidney injury in the index case were deemed multifactorial. However, a multidisciplinary team concluded that the primary cause was silicone-induced granulomatous disease, as they occurred with the development of granulomatous mediastinal lymphadenopathy after silicone exposure during difficult breast implant extraction, and her parathyroid hormone levels were suppressed.

The patient was commenced on oral prednisolone (initial dose of 0.5 mg/kg) and was advised to modify her diet and limit sunlight exposure. During a review 3 weeks later, she reported significant improvement in symptoms, and serum calcium and creatinine levels were normalized (Figure 4). However, her symptomatic hypercalcemia relapsed on discontinuing systemic steroids, even until 3 months after implant removal. The reintroduction of a low dose of prednisolone (10 mg daily) resolved hypercalcemia and controlled her symptoms. She was counseled about the likely protracted course of illness, alternate or steroid-sparing therapeutic options, and other potential complications, including anaplastic large-cell lymphoma.

Discussion

Hypercalcemia is typically caused by primary hyperparathyroidism or malignancy and also can be associated with granulomatous diseases of various etiologies, including sarcoidosis and tuberculosis. Hypercalcemia secondary to silicone exposure in topical (4) and injection forms (5, 6) has been previously published. However, breast implant rupture–related symptomatic hypercalcemia has not yet been widely documented. To the best of our knowledge, this case is only the fourth-reported instance of symptomatic hypercalcemia resulting from silicone breast implant rupture–induced

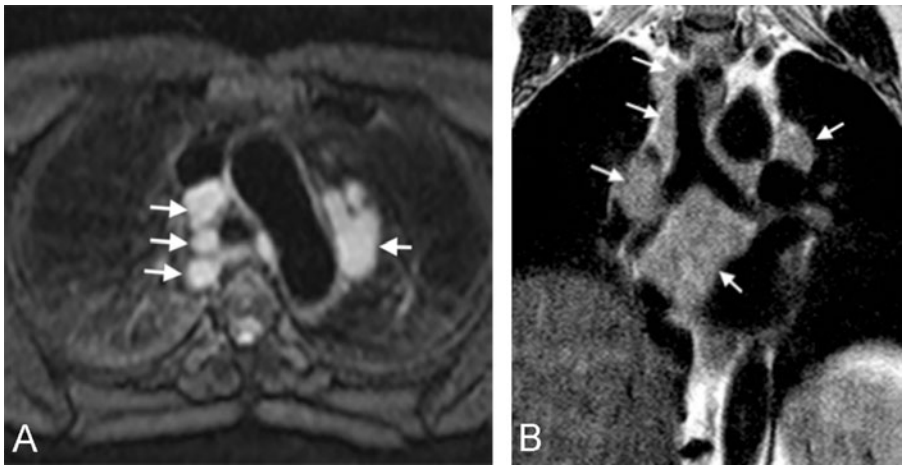

Figure 2. (A) MRI, axial short tau inversion recovery view, shows enlarged mediastinal and hilar lymph nodes (*arrows*). (B) MRI T2-weighted coronal view shows enlarged mediastinal, hilar and subcarinal lymph nodes (*arrows*). MRI = magnetic resonance imaging.

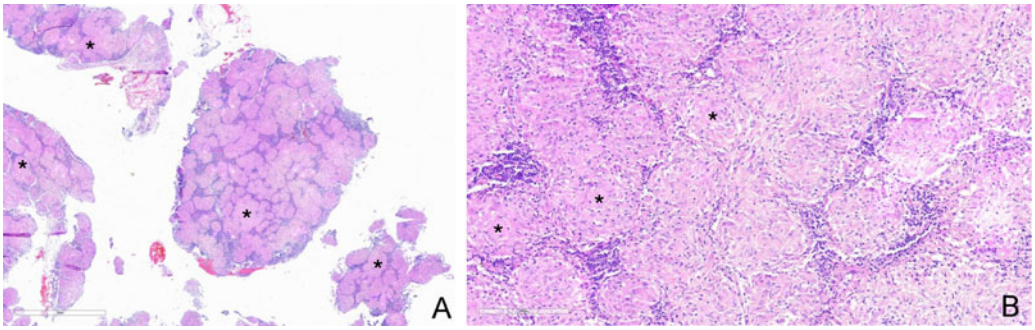

Figure 3. **(A) Mediastinal lymph node biopsy (×20 magnification) shows fragments of nodal tissue with well-formed noncaseating granulomas (*asterisks*). (B) Mediastinal lymph node biopsy (×200 magnification) shows nodal tissue with well-formed noncaseating granulomas (*asterisks*).**

granulomatous disease (7, 8) and the second case of prolonged or refractory hypercalcemia due to this cause (8). Although some previous publications have suggested that silicone breast implant–induced granulomas may be a form of sarcoidosis (9–11), this diagnosis should only be considered after ruling out the known causes of granulomatous disease (12).

Breast implant rupture in our case was mostly due to their prolonged age (35 years). Unfortunately, the patient did not consider periodic follow-up imaging since augmentation, as recommended by the Food and Drug Administration (2). Silicone particles from her older noncohesive implants might have migrated during difficult surgical removal, leading to an intense granulomatous reaction in the intrathoracic lymph nodes. Hypercalcemia in the index case likely resulted from increased intestinal calcium absorption and mobilization from bones due to extrarenal calcitriol production by lymph node macrophages, which is resistant to normal parathyroid hormone feedback control. Parathyroid hormone–related protein also has been reported to contribute to hypercalcemia in granulomatous diseases (13). A limitation of the present case is the lack of measurement of serum vitamin D levels. Although an elevated 1,25-dihydroxy vitamin D level could have further supported the underlying pathogenesis. Nevertheless, the diagnosis in this case is firmly established by the sequence of events, including the onset of acute hypercalcemia concurrent with extensive granulomatous disease following silicone exposure during breast implant removal.

Granulomatous disease associated hypercalcemia responds well to systemic steroids, which act by inhibiting calcitriol synthesis by the activated mononuclear cells (14). However, steroids may not be as effective in treating hypercalcemia caused by silicone exposure (8). In addition, the effectiveness of alternative therapies such as bisphosphonates, ketoconazole, infliximab, and hydroxychloroquine for this condition is unknown. The long-term course and sequelae of hypercalcemia related to silicone breast implant rupture–induced granulomatous disease remain uncertain. This patient's case underscores the necessity of monitoring silicone breast implants with periodic magnetic resonance imaging and being vigilant for complications that may arise from their rupture.

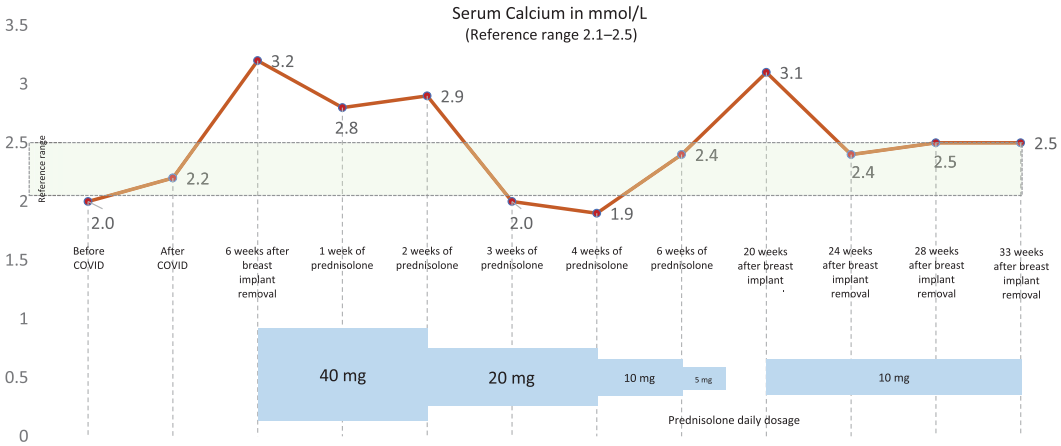

Figure 4. **Line graph displaying the progression of serum calcium levels before and after breast implant removal, as well as response to oral prednisolone therapy (reference range 2.1–2.5 mmol/L).**

References

1. American Society of Plastic Surgeons (ASPS). 2018 plastic surgery statistics report. 2018. Accessed at https://www.plasticsurgery.org/documents/News/Statistics/2018/plastic-surgery-statistics-full-report-2018.pdf on 26 February 2023.
2. The US Food and Drug Administration (FDA). Risks and complications of breast implants. 2022. Accessed at https://www.fda.gov/medical-devices/breast-implants/risks-and-complications-breast-implants. on 26 February 2023
3. Safvi A. Linguine sign. Radiology. 2000;216:838-9. [PMID: 10966719] doi:10.1148/radiology.216.3.r00se16838
4. Schanz J, Flux K, Kircher C, et al. Mirror, mirror on the wall: hypercalcemia as a consequence of modern cosmetic treatment with liquid silicone. Med Sci Monit. 2012;18:CS5-7. [PMID: 22293885] doi:10.12659/msm.882450.
5. Kozeny GA, Barbato AL, Bansal VK, et al. Hypercalcemia associated with silicone-induced granulomas. N Engl J Med. 1984;311:1103-5. [PMID: 6548295] doi:10.1056/NEJM198410253111707.
6. Tachamo N, Donato A, Timilsina B, et al. Hypercalcemia associated with cosmetic injections: a systematic review. Eur J Endocrinol. 2018;178:425-30. [PMID: 29453201] doi:10.1530/EJE-17-0938.
7. Rodriguez IE, Deleyiannis FW. Hypercalcemia secondary to silicone breast implant rupture: a rare entity to keep in mind. Plast Reconstr Surg Glob Open. 2017;5:e1416. [PMID: 28831356] doi:10.1097/GOX.0000000000001416.
8. Dangol GMS, Negrete H. Silicone-induced granulomatous reaction causing severe hypercalcemia: case report and literature review. Case Rep Nephrol. 2019;2019:9126172. [PMID: 30729052] doi:10.1155/2019/9126172.
9. Watad A, Rosenberg V, Tiosano S, et al. Silicone breast implants and the risk for autoimmune/rheumatic disorders: a real-world analysis. Int J Epidemiol. 2018;47:1846-54. [PMID: 30329056] doi:10.1093/ije/dyy217
10. Gavoille A, Vincent M, Kiakouama L, et al. Sarcoidosis after breast implant rupture: looking beyond granulomas. Autoimmun Rev. 2020;19:102673. [PMID: 32942033] doi:10.1016/j.autrev.2020.102673
11. Todorov TI, de Bakker E, Smith D, et al. A case of silicone and sarcoid granulomas in a patient with “highly cohesive” silicone breast implants: a histopathologic and laser Raman microprobe analysis. Int J Environ Res Public Health. 2021;18:4526. [PMID: 33923240] doi:10.3390/ijerph18094526.
12. Crouser ED, Maier LA, Wilson KC, et al. Diagnosis and detection of sarcoidosis. An official American Thoracic Society Clinical Practice Guideline. Am J Respir Crit Care Med. 2020;201:e26-51. [PMID: 32293205] doi:10.1164/rccm.202002-0251ST.
13. Zeimer HJ, Greenaway TM, Slavin J, et al. Parathyroid-hormone-related protein in sarcoidosis. Am J Pathol. 1998;152:17-21. [PMID: 9422518]
14. Sandler LM, Winearls CG, Fraher LJ, et al. Studies of the hypercalcaemia of sarcoidosis: effect of steroids and exogenous vitamin D3 on the circulating concentrations of 1,25-dihydroxy vitamin D3. Q J Med. 1984;53:165-80. [PMID: 6087399]