

A Severe Case of Siliconoma-induced Hypercalcemia due to Illicit Gluteal Silicone Injections

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Summary: Siliconoma-induced hypercalcemia is a rare complication of siliconoma, occurring secondary to a foreign body granulomatous process induced by the introduction of silicone into soft tissue. This is a case report of a woman presenting with sequelae of illicit silicone injections performed in an unknown woman's basement in Florida 20 years before presentation. A 39-year-old woman presented with a 2-month history of 20-pound weight loss, malaise, and intractable vomiting with a remote history of unregulated cosmetic injections to the bilateral gluteal and thigh regions. Her laboratory studies were consistent with severe hypercalcemia secondary to a foreign body granulomatous process. Initially, she was medically managed, with mild improvements in her symptomatic hypercalcemia and later underwent palliative debridement with siliconoma removal. Postoperatively, her course was complicated by delayed wound healing and graft failure, but the surgical defect was later closed successfully with split-thickness skin grafting after months of wound care. Although the procedure was not intended to treat her hypercalcemia, there were significant improvements in serum and ionized calcium in the months following her procedure. Severe hypercalcemia in the context of previous unregulated cosmetic injections or possible silicone implant rupture should prompt consideration of siliconoma-induced hypercalcemia as the underlying etiology. In addition to the established utility of IV fluids, bisphosphonates, and glucocorticoids, there may also be a role for surgical intervention in the management of this unique patient population's hypercalcemia. (*Plast Reconstr Surg Glob Open* 2022;10:e4184; doi: [10.1097/GOX.0000000000004184](https://doi.org/10.1097/GOX.0000000000004184); Published online 14 March 2022.)

Historically, cosmetic liquid silicone injectables (LSI) gained popularity during World War II, becoming increasingly prevalent in the United States until soft tissue complications precipitated their ban in 1975.^{1,2} LSI are not FDA-approved for cosmetic use; however, “black market” injections are sought out as cheaper alternatives to FDA-approved cosmetic fillers. Further, patients participate in cosmetic tourism to circumvent protective restrictions in the United States to inexpensively obtain enhancement abroad.³ Most concerning is that individuals administering LSI are often untrained, unqualified individuals who deceive patients by presenting themselves as medical professionals.⁴

Societally, LSI are discussed through criminal investigations as practitioners are prosecuted or through celebrity stories, such as Cardi B's infamous GQ interview.^{1,5} Investigations require years in development due to the latent period between initial injection and manifestation of complications.^{1,6} Although the number of cosmetic procedures performed by unlicensed individuals is unknown, a 2013 study identified 28 cases through news articles and lawsuits, indicating geographic predominance in Florida (35.7%), Texas (14.3%), and California (10.7%).⁶ The FDA and ASPS have released warnings about unregulated injections and procedures to deter patients from seeking out these potentially lethal procedures.^{1,4,5}

CASE DESCRIPTION

A 39-year-old woman who underwent silicone injections to her bilateral thigh and gluteal regions 20 years prior presented with malaise, intractable vomiting, and a 2-month history of 20-pound weight loss. She appeared cachectic with laboratory studies showing elevated serum

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Table 1. Table of Laboratory Test Results at Presentation and following Discharge with Listed Reference Values

Laboratory Test	Presentation	Following Discharge	Reference
	Dec 2020	May 2021	
Serum calcium (mg/dL)	17.0	10.2	8.5–10.1
Direct ionized calcium (mmol/L)	1.88	1.35	1.18–1.32
Parathyroid hormone (pg/mL)	<6.3	<6.3	18.5–88.0
Vitamin D 1,25-DI-OH (pg/mL)	232	81.3	19.9–79.3
Vitamin D, 25-OH (ng/mL)	58	22	30–100
Parathyroid-related peptide (pmol/L)	11.3	8.2	0.0–3.4
Angiotensin converting enzyme (U/L)	77	—	9–67
Serum phosphate (mg/dL)	3.1	4.1	2.3–4.6
Alkaline phosphatase (U/L)	39	90	35–120
Thyroid stimulating hormone (μU/mL)	3.77	3.90	0.36–3.74

calcium of 17.0 (ref. 8.5–10.1 mg/dL), elevated calcitriol of 232 pg/mL (ref. 19.9–79.3 pg/mL), and decreased PTH of less than 6.3 (ref. 18.5–88.0 pg/mL) (Table 1). The etiology was initially unclear. Siliconoma-induced hypercalcemia (SIH) was a diagnosis of exclusion after evaluating for malignancy, autoimmune disease, paraneoplastic syndromes, and granulomatous diseases with extensive workup, including imaging, bone marrow biopsy, and rheumatologic workup. SIH was supported by her history of gluteal injections, laboratory evidence of PTH-independent calcitriol-mediated hypercalcemia (Table 1), multiple granulomata on magnetic resonance imaging (MRI) consistent with siliconoma, and otherwise nondiagnostic evaluation (see Fig. 1).

The diagnosis was later confirmed with gold standard tissue biopsy showing foreign-body granulomata.

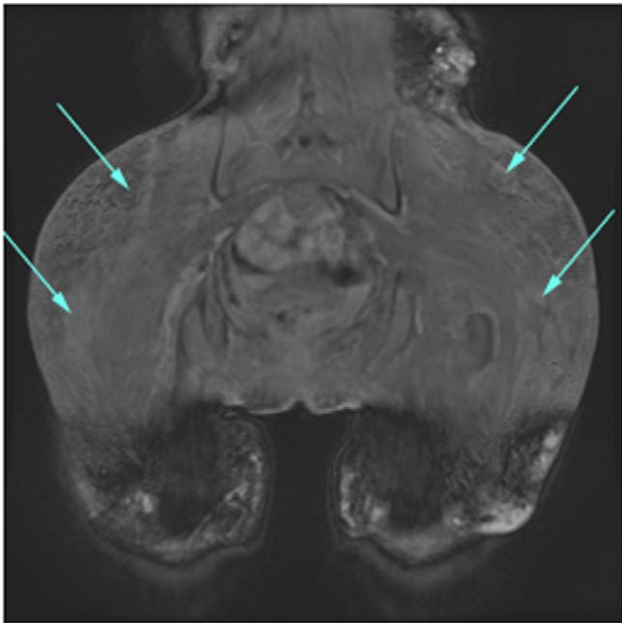


Fig. 1. MRI of the pelvis with 4.5 cc Gadavist intravenous contrast. Coronal view of the pelvis exhibiting extensive subcutaneous edema and multiple foreign body granuloma penetrating to the depth of superficial muscle and scattered throughout the gluteal region, lower anterior pelvis, and proximal thighs with prominent inguinal lymph nodes. Arrows indicate foreign body granulomatous infiltration.

Throughout her longitudinal course of care, her hypercalcemia was managed medically with continuous intravenous hydration and varying courses of bisphosphonates, corticosteroids, and calcitonin (Fig. 2). She experienced intermittent mild gluteal and thigh pain since the initial injections due to tissue hardening and inflammation; however, 3 months preceding admission, her pain gradually worsened with associated muscle weakness without paresthesia or neurologic deficit. Due to debilitating pain limiting her ability to walk, palliative debridement was offered. Surgery was not expected to affect her hypercalcemia and significant morbidity was expected given the extent of tissue involvement. The patient and her family decided to proceed with surgery. The bilateral gluteal and thigh regions were extensively debrided, resecting subcutaneous tissue, necrotic fascia, and abutting superficial muscle without deep muscular invasion (Fig. 3).

A week later, she returned to the OR for additional debridement and split-thickness skin grafting with negative pressure wound therapy (NPWT). Although the tissue bed appeared well-granulated and noninfected, the grafts did not take. The failed graft was removed, and she was maintained on NPWT. Modest improvements in peak serum and ionized calcium were observed postoperatively (Table 1, Fig. 1). After 3 months of wound care, she underwent the final debridement with repeat split-thickness skin grafting. Postoperatively, she was managed with NPWT with excellent wound healing and graft uptake. (See figure 1, Supplemental Digital Content 1, which demonstrates wound progression over time. <http://links.lww.com/PRSGO/B965>.)

Seven months following her final procedure, she is doing extremely well on outpatient follow-up. Her wounds have healed, and her nutritional status has improved substantially. Her serum and ionized calcium continue to normalize. She is returning to work, and her quality of life has dramatically improved from her initial presentation.

DISCUSSION/CONCLUSION

Acutely following LSI administration, swelling and pain may occur at the injection site, but it can take up to 28 years for sequelae such as hyperpigmentation, necrosis, and granuloma to develop.^{1,2,7,8} Systemically, granuloma formation secondary to silicone, termed siliconoma, rarely causes hypercalcemia through disordered production of 1,25-dihydroxycholecalciferol by activated macrophages similar to sarcoidosis.^{8–10} Elevated calcitriol and serum calcium with suppressed PTH are hallmark laboratory findings. Through a cytokine-dependent inflammatory pathway, PTHrP may also be elevated.¹⁰ Patients with SIH present with nonspecific symptoms of hypercalcemia, including fatigue, abdominal pain, cognitive decline, and weight loss in the context of previous cosmetic injections.⁹ Guidelines for SIH are not standardized due to few reported cases and insufficient data supporting a definitive cure.^{1,9} Consensus supports managing the hypercalcemia with IV hydration, corticosteroids, and bisphosphonates.^{1,8}

Siliconoma resection is challenging, given unclear margins with widespread silicone infiltration, making

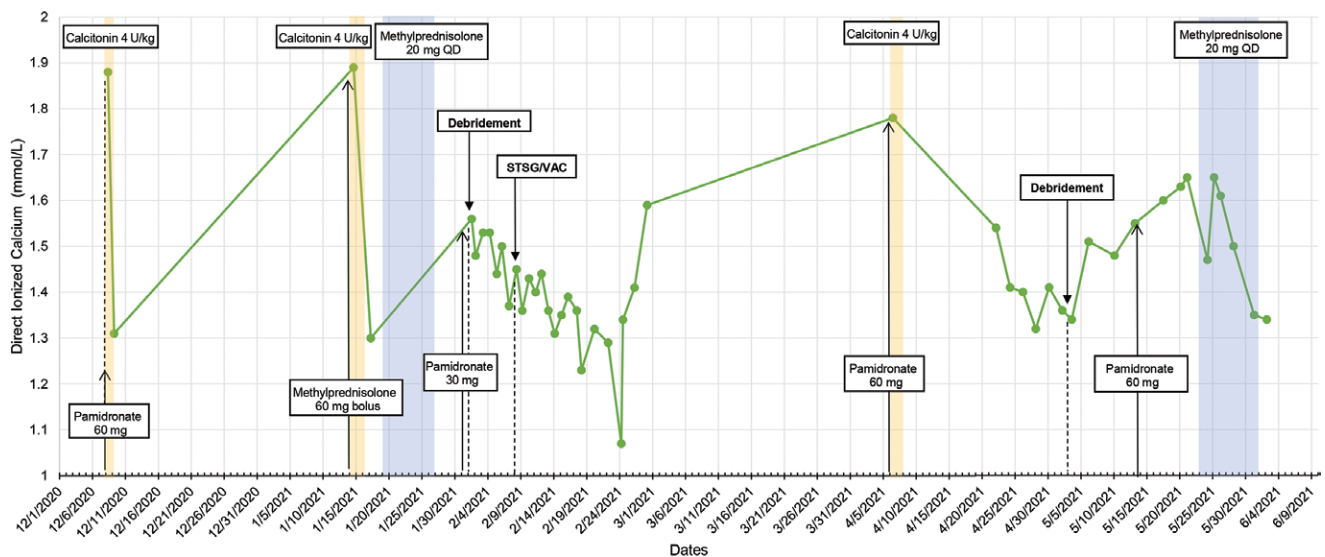


Fig. 2. Trend of direct ionized calcium with superimposed timeline of medical and surgical management as a longitudinal trend over course of care.

complete eradication of diseased tissue impossible.^{1,7,8} Substantial debridement is typically required, resulting in poor cosmesis.^{1,7} Closure is similarly difficult, as delayed wound healing is likely.^{2,7} The indications for siliconeoma resection are severe manifestations, including



Fig. 3. Image showing wound appearance of left gluteal region on postoperative day 2 following the first debridement.

epidermolysis, abscess, fistula, and necrosis or insufficient improvement with medical management.¹ Surgical indications in SIH are less defined.^{7,8} However, severe tissue manifestations and recalcitrant hypercalcemia are reasonable indications for debridement.⁷

For our patient, surgery was intended to be palliative. However, her laboratory abnormalities gradually improved with decreases in calcitriol (Table 1), serum calcium (Table 1), and direct ionized calcium (Fig. 2). Interestingly, after initial debridement, a negative association was observed between direct serum calcium and time over the 30-day postoperative period despite an otherwise stable treatment regimen (Fig. 1). A Pearson's correlation coefficient was computed to assess the relationship, showing a statistically-significant moderate negative linear association ($r(22) = -0.5001$, $P = 0.012825$) (See graph, Supplemental Digital Content 2, which shows the negative linear association of direct ionized calcium over the 30-day postoperative period. <http://links.lww.com/PRSGO/B966>).

While appearing to be temporally associated, our report is insufficient to establish causation due to inherent limitations of single patient studies as well as concomitant medical management. Further, we hypothesize that the striking decline in direct ionized calcium was only seen after the initial surgery because the majority of abnormal tissue was removed at this time. Considerably less tissue was removed in the two subsequent surgeries. This indicates that complete resection may not be required, but rather that a critical mass of tissue might require debulking to see therapeutic benefits.

In this article, we present a case of SIH incited by illegal silicone injections performed 20 years before presentation, and we characterize an association with hypercalcemia improvement in the postoperative period. Although our study adds to the literature describing a positive outcome in SIH following debridement, additional research is needed to elucidate the specific utility of surgery in this patient

population. By highlighting this case, we hope to increase awareness of the dangers of LSI and inform the future treatment of patients presenting with this rare disease entity.

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