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Case Presentation

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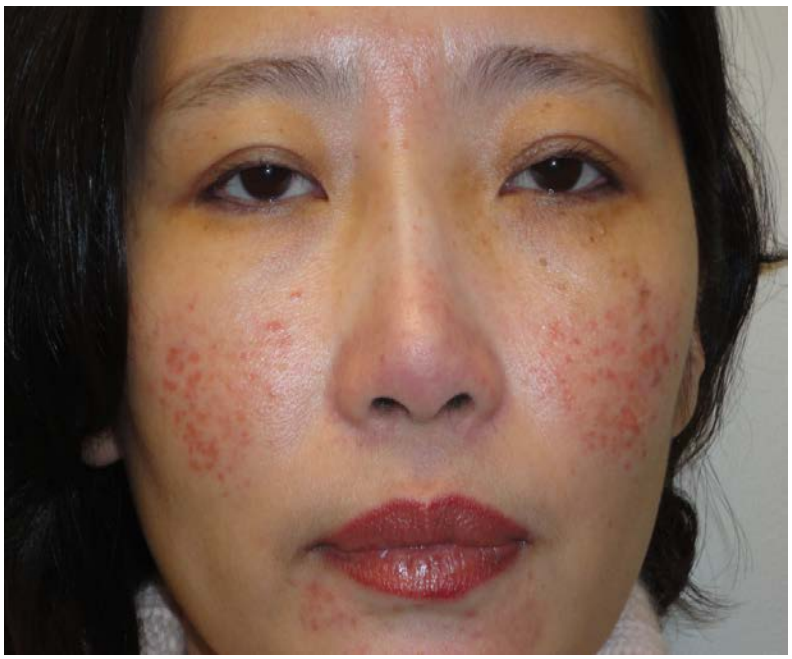
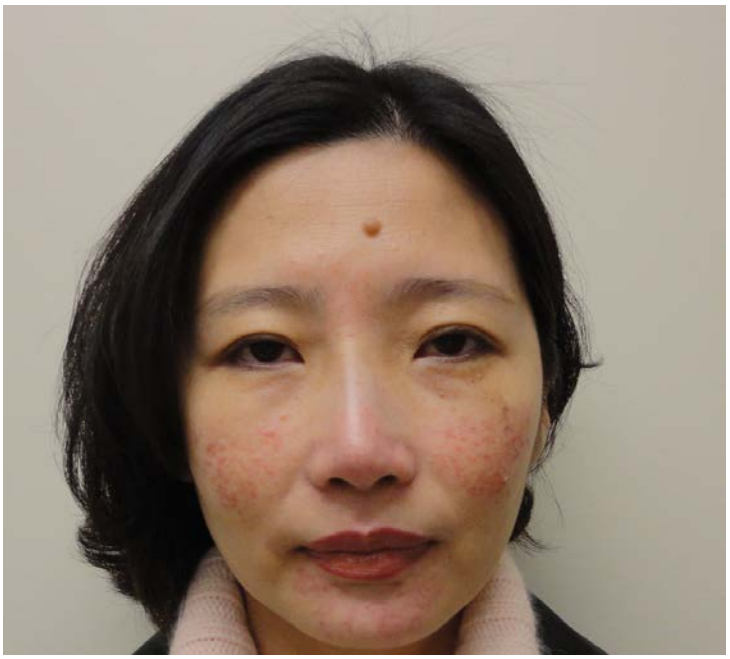
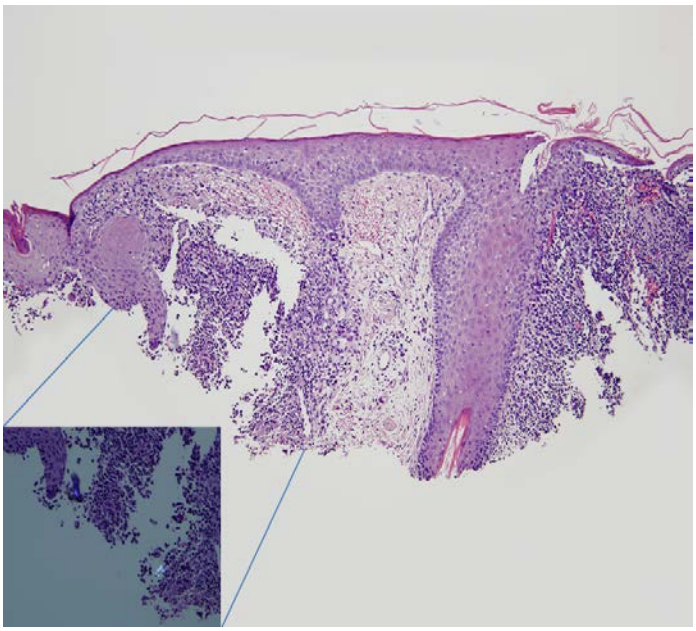
Abstract

Silicone in liquid and gel implantation may induce granuloma formation and migration. Although there are many complications associated with solid silicone implantation, there have been no published reports of distant granuloma formation. We present a case of a woman with clinical and serologic findings that are consistent with systemic lupus erythematosus and a histopathologic diagnosis of foreign body granulomatous dermatitis 20 years after solid silicone nasal implantation. We review the literature on silicone granulomas and their treatment and speculate on the potential etiologies of a challenging case presentation.







Case synopsis

A 51-year-old woman presented to the Skin and Cancer Unit in September, 2012, with a two-year history of facial redness. She had no local symptoms but did have intermittent arthralgias, Raynaud's phenomenon, and morning swelling of the fingers. Her past medical history included glaucoma, cholecystectomy, a solid silicone nose implantation 20 years prior to presentation, and a breast lumpectomy. She denied fevers, chills, weight loss, nocturnal diaphoresis, sun sensitivity, fatigue, chest pain, and shortness of breath. Prior to her presentation she had been treated for presumed rosacea but prematurely terminated a trial of doxycycline owing to gastrointestinal side effects. She was later treated for suspected systemic lupus erythematosus with hydroxychloroquine 200 mg daily but stopped after two months without improvement. She then underwent intralesional glucocorticoid injections and pulsed-dye laser therapy without improvement.

Physical Examination: Erythematous papules, some with fine overlying scale, were present on the malar eminences. There was erythema at the nasal tip.

Laboratory Data: A complete blood count and comprehensive metabolic panel were normal. Anti-nuclear antibody was positive at 1:160, with a homogenous, speckled pattern. C3 was low at 81mg/dL. C4 was normal. Anti-Smith, anti-ribonucleoprotein, anti-Ro/SSA, and anti-La/SSB cap antibodies were negative.

Histopathology: There is a perivascular and perifollicular, mixed-cell inflammatory infiltrate with lymphocytes, histiocytes, plasma cells, neutrophils, and scattered eosinophils. Multinucleated giant cells are identified, and there is focal polarizable foreign material.

Diagnosis: Granulomatous dermatitis

Discussion: This patient is a very challenging case because she presented with a distribution of lesions, associated systemic symptoms, and serologic markers that were consistent with systemic lupus erythematosus (SLE), but histopathologic findings of foreign-body granulomatous dermatitis. Owing to her history of silicone nose implantation, our clinical consensus is that, in addition to having SLE, she developed a granulomatous response to a component of her nasal implant. Our discussion will briefly review the uses and complications of silicone and offer a potential explanation for this stimulating case.

Silicone (polysiloxane) is known widely by general, cosmetic, and surgical dermatologists and is available in gel-filled implants as well as in liquid and solid forms. Liquid and gel implants may be used for tissue augmentation (e.g breast) and as an aesthetic filler (e.g. in HIV-associated lipoatrophy) [1]. In some patients the use of gel and liquid injections has been followed, usually months to years afterwards, by the formation of cutaneous nodules and plaques. In cases of silicone implant rupture, gel has leaked from the implant and spread to distant sites such as the abdominal wall and arms. Similar migrations have been observed after procedures that involve injectable silicone [1, 2]. One particularly striking case identified metastatic silicone granulomas in the form of lupus miliaris disseminatus faciei-like facial nodules in a silicone breast implant recipient [3]. Although there have been anecdotal reports of patients developing autoimmune diseases after silicone breast augmentation, several large epidemiologic studies have unequivocally refuted this association [1, 4]. Still a controversial subject, silicone (in any form) may provide excellent results when utilized by an experienced practitioner, but to avoid numerous potential adverse events, its use requires meticulous technique.

Solid silicone rubber implantation is the most popular rhinoplasty procedure in East Asia. Its advantages include low cost, a stable chemical structure, and a low degree of tissue reactivity that allows for ease of molding to the desired contour [5]. However, as with the injectable and gel forms, solid silicone acts as a foreign body and induces the host response of fibrous encapsulation rather than tissue integration [6, 7]. Complications often depend on the individual host and the location of implant insertion, with subcutaneous and subfascial implantations causing greater numbers of complications than deeper, subperiosteal implantations [8]. Complications that are associated with solid implantations have included protrusion, infection, graft displacement, dyspigmentation, dysesthesia, and graft drift [7-10]. There have been no reported instances of solid implant rupture or granulomatous nodule formation distant from the site of implantation. There have been no reports of incident or reactivated connective-tissue disease after silicone nose implant.

The pathogenesis of silicone granuloma formation and migration are unknown. Multiple theories attempt to explain the development of granulomas. These include infectious events or systemic inflammatory processes at distant sites as a potential link and injectable silicone particulate matter as a nidus for infection in the form of bacterial biofilm [11]. In the latter theory, a low-grade, chronic infection could persist for years before presenting as a granuloma [12]. In addition, because silicone is incorporated into the reticuloendothelial system, it may be thought of as eliciting a foreign-body granulomatous response [13, 14]. Silicone migration is not well understood but may be location-dependent and related to the amount of silicone injected or implanted [14].

Treatment of foreign-body granuloma can be challenging. Surgical excision may not offer an acceptable cosmetic result and may not be possible when migration has occurred. Intralesional and oral glucocorticoids may temporarily improve lesions, although relapses are common when treatment is discontinued. Allopurinol, imiquimod, minocycline, and low-dose oral retinoids also have been reported to be helpful in the treatment of silicone granuloma in case reports [15, 16]. Treatment of this patient was particularly challenging because of her poor compliance. She prematurely discontinued multiple medications without completing a course of any. Attempted treatments included mycophenolate mofetil, azathioprine, triamcinolone cream, and clobetasol cream. She is currently undergoing a trial of low-dose isotretinoin.

Another challenge was the fact that the exact composition of the silicone nose implant and operative report were not available. Because the histopathologic features did not demonstrate the classic swiss cheese pattern of silicone granuloma, it is likely that the reaction identified was owing to the presence of another foreign material, perhaps an additive of an adulterated implant or a contaminant of a procedure performed under unsterile conditions. Because of its low cost and easy availability as an industrial substance, silicone, especially in liquid form, has been extensively altered, substituted, and misused by untrained providers [13]. It is thus possible that the patient's implant may not have been of medical quality, may not have been purely solid in form, and may have ruptured, thus leaking its contents and stimulating a granulomatous response. It is also plausible that an additional foreign substance was introduced during the implantation procedure. Finally, although the association between silicone and connective tissue diseases has been thoroughly refuted via meta-analysis, there still remain numerous case reports of the development of connective tissue disease after procedures that involve the use of silicone. Objective data have demonstrated that patients with immune-mediated reactions to silicone implants had increased IgG in surrounding tissue when compared with asymptomatic implanted patients [17]. In addition, serum anti-silicone antibodies have been more frequently detected in patients with silicone implants when compared with controls [18]. Moreover mice that spontaneously developed autoimmune diseases after silicone implantation have demonstrated elevations in various autoantibodies [19]. Although speculative, it seems plausible that a subgroup of silicone implanted patients may be at risk of developing autoimmune diseases [20].

Owing to the strong evidence base refuting the association between silicone and connective-tissue diseases, it seems most likely that our patient had two distinct, coincidental, and unrelated disease entities, SLE and foreign-body granulomatous dermatitis. However, it is theoretically possible that our patient represents an atypical case in which foreign material did induce or reactivate a connective tissue disorder. We are unable to make a firmer conclusion because of the limitations of the chronology of the patient's clinical course prior to presentation and the lack of laboratory data prior to nasal implantation.

Our patient presents an atypical and challenging case of foreign-body granuloma after solid silicone nose implantation. To our knowledge, this case is the first to report distant spread of granulomas after solid silicone nasal implant rhinoplasty.

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