

Siliconoma successfully controlled with low-dose oral isotretinoin: A case report with histopathologic and ultrasonographic findings



Siriwan Palawisuth, MD,^{a,b} Janice Natasha C. Ng, MD,^a Penvadee Pattanaprichakul, MD,^a Yanisorn Nanchaipruek, MD,^a Nuttagarn Jantanapornchai, MD,^a Rungsima Wanitphakdeedecha, MD, MSc,^a and Sasima Eimpunth, MD^a
Bangkok and Nonthaburi, Thailand

Key words: foreign body granuloma; injectable silicone granuloma; isotretinoin; siliconoma.

INTRODUCTION

Liquid injectable silicone (LIS) has been used for soft tissue augmentation for more than 5 decades; however, its use remains controversial due to associated complications.¹ Granulomatous reaction or siliconoma is the most commonly observed complication, and it clinically presents as recurrent cellulitis, skin induration, nodules, ulceration, and/or local lymph node involvement.² Treatment of siliconoma remains a therapeutic challenge. Here, we present the case of a 65-year-old woman with facial siliconoma who responded well to low-dose isotretinoin.

CASE REPORT

A 65-year-old Thai woman was referred to the Department of Dermatology of the Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand with a 2-week history of swelling of both cheeks. Her history revealed that she had undergone injection of an unknown substance into her cheeks approximately 30 years ago for cosmetic enhancement. She also had a history of chronic hepatitis C (genotype 1a) infection with liver cirrhosis, which responded well to ledipasvir/sofosbuvir (Ledvir; Mylan Laboratories Limited) 90 mg/400 mg and ribavirin (Copegus; Roche).

Physical examination revealed diffuse, indurated, firm, and fixed plaques on both cheeks (Fig 1, A). An incisional biopsy taken from the left cheek revealed siliconoma with foreign-body

Abbreviation used:

LIS: liquid injectable silicone

granulomatous reaction (Fig 2). Acid-fast bacilli stain, Gomori methenamine silver stain, and periodic acid-Schiff with diastase stain were negative. Bacteriologic culture, mycologic culture, mycobacterium culture, and direct immunofluorescence were also negative. Complete blood cell count, liver function tests, blood chemistry, and lipid profile were all within normal limits.

Prednisolone 20 mg/day and loratadine 10 mg/day were initially prescribed, and after 2 weeks of treatment, the patient reported some improvement. However, considering the risks associated with long-term systemic corticosteroid use (especially during the COVID-19 pandemic), other treatment options were discussed with the patient. Isotretinoin 10 mg/day (5 tablets/week) was then started. After 3 months, there was dramatic improvement in the induration and swelling at both cheeks, as shown in Fig 1, C. The isotretinoin dose was gradually tapered to 40 mg/week for 1 month, after which the dose was maintained at 30 mg/week for 7 months combined with oral prednisolone at 2.5-5 mg/day. Repeat liver function tests at 2 and 6 months after the beginning of the oral isotretinoin treatment were normal. The patient reported no recurrence of swelling since the

From the Department of Dermatology, Faculty of Medicine Siriraj Hospital, Mahidol University^a and Department of Medicine, Panyanantaphikkhu Chonprathan Medical Center, Srinakharinwirot University.^b

Funding sources: None.

IRB approval status: Not applicable.

Correspondence to: Sasima Eimpunth, MD, Associate Professor of Dermatology, Department of Dermatology, Faculty of Medicine Siriraj Hospital, Mahidol University, 2 Wanglang Road, Bangkok 10700, Thailand. E-mail: doctorsasima@gmail.com.

JAAD Case Reports 2021;14:24-6.

2352-5126

© 2021 by the American Academy of Dermatology, Inc. Published by Elsevier, Inc. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

<https://doi.org/10.1016/j.jdc.2021.06.001>



Fig 1. Clinical photography at baseline (A), 1 month after initiation of prednisolone treatment (B), 3 months after initiation of isotretinoin treatment (C), and 6 months after initiation of isotretinoin treatment (D).

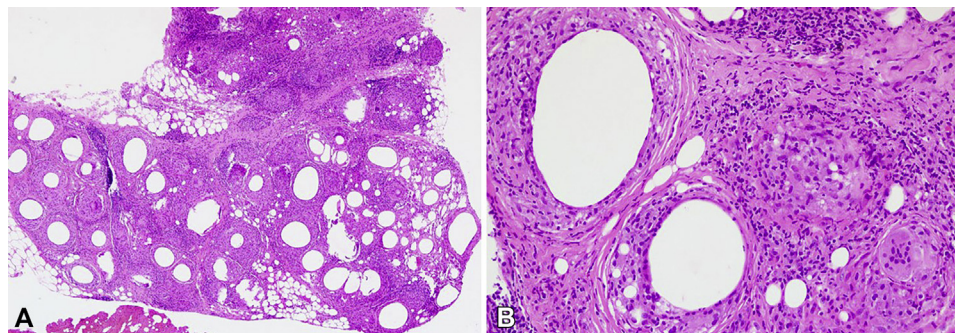


Fig 2. Diffuse dermal and subcutaneous infiltration by clear cystic spaces of varying sizes surrounded by foreign-body granulomatous inflammation (A and B, Hematoxylin-eosin stain; original magnifications: A, $\times 40$; B, $\times 400$).

beginning of the treatment; however, multiple small indurated nontender plaques were still present on both cheeks at the 6-month follow-up (Fig 1, D). Ultrasonographic findings compared between non-indurated normal skin and indurated skins at the left cheek are presented in Fig 3. The indurated area shows a well-defined cystic lesion, which is a typical finding in siliconoma.³

DISCUSSION

Silicone is a synthetic compound composed of long polymers of dimethylsiloxanes.² It is permanent, nontoxic, noncarcinogenic, inert, and minimally antigenic. Silicone is available in liquid and

solid forms, and LIS is mainly used for soft tissue augmentation.⁴ LIS can persist in tissue because it is composed of nonbiodegradable molecules. The small droplets will be phagocytosed by macrophages into microdroplets, whereas larger volumes will migrate along the tissue planes.⁵ Complications associated with LIS include edema, pain, ecchymosis, erythema, pigmentation, and embolism if injected into the vascular system.⁶ Other severe complications of LIS include acute pneumonitis,² granulomatous hepatitis,² and hypercalcemia associated with siliconoma.⁷ Inflammation can occur months to years after injection. Some theories have been proposed to explain LIS-induced

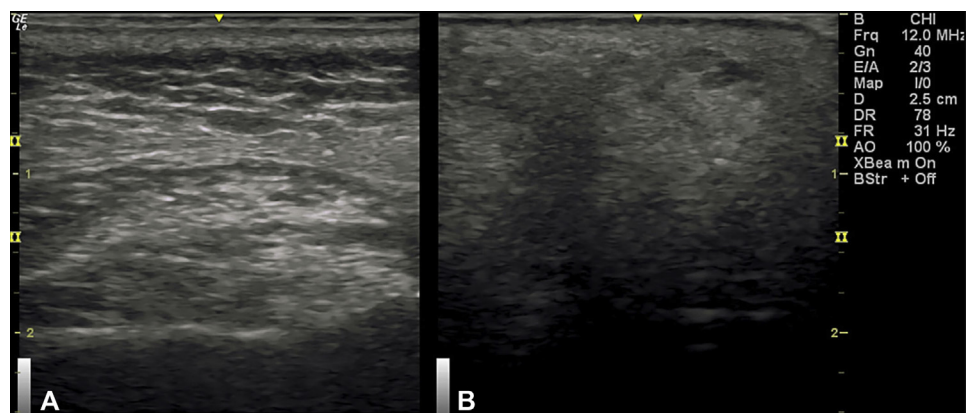


Fig 3. Ultrasonographic findings in the normal nonindurated skin (A), and in indurated skin at 6 months after the start of isotretinoin treatment (B).

inflammation, including immunologic viral cross-reactivity and acute and/or chronic inflammation due to silicone impurity. Monocytes are differentiated into macrophages, which play a role in the release of cytokines, such as interferon gamma and tumor necrosis factor alpha, during the granulomatous response.⁸

A diagnosis of siliconoma can be made by histopathologic confirmation. In order to determine the dimension or boundary of the granuloma for treatment planning or follow-up, many types of imaging studies can be performed, such as ultrasonography, computed tomography, and/or magnetic resonance imaging.⁸ In our case, definite diagnosis was confirmed by histopathology with negative culture and negative direct immunofluorescence.

The treatment options for localized granuloma include systemic and local corticosteroids, 5-fluorouracil,⁸ allopurinol,⁴ low-dose isotretinoin (20 mg/day over 6 months),^{8,9} methotrexate,¹⁰ antibiotics, and surgical resection.⁴ Antibiotics, especially minocycline⁹ and doxycycline,⁹ have been used successfully. Additional treatment modalities that directly target the granuloma include etanercept (tumor necrosis factor alpha inhibitor); imiquimod (Toll-like receptor activator with immune antiproliferative effects), which works by stimulating both the innate and cell-mediated immune system via interferon gamma²; and tacrolimus (macrolide immunosuppressant).⁸

In our case, the patient gradually recovered within 3 months after beginning low-dose oral isotretinoin treatment. Medication therapy was preferred over surgical intervention due to extensive involvement of both cheeks. Isotretinoin was considered because of its anti-inflammatory properties.⁸ Considerable improvement was achieved over a relatively short

period of time. The results reported here suggest the need for further investigation of isotretinoin as a treatment for siliconoma.

The authors gratefully acknowledge the patient profiled in this report for permitting us to disclose details relating to her case. The patient in this manuscript has given written informed consent to publication of the case details.

Conflicts of interest

None declared.

REFERENCES

1. Ellis LZ, Cohen JL, High W. Granulomatous reaction to silicone injection. *J Clin Aesthet Dermatol*. 2012;5(7):44-47.
2. Baumann LS, Halem ML. Lip silicone granulomatous foreign body reaction treated with Aldara (imiquimod 5%). *Dermatol Surg*. 2003;29(4):429-432.
3. Ali L, McGivern D, Teoh R. Silicon granuloma mimicking lung cancer. *BMJ Case Rep*. 2012;2012:bcr2012006351.
4. Chen YC, Chen ML, Chiu YM. A case of mimicking angioedema: chin silicone granulomatous reaction spreading all over the face after receiving liquid silicone injection forty years previously. *Chin Med J (Engl)*. 2011;124(11):1747-1750.
5. Harlim A, Kanoko M, Aisah S. Classification of foreign body reactions due to industrial silicone injection. *Dermatol Surg*. 2018;44(9):1174-1182.
6. Rapaport MJ, Vinnik C, Zarem H. Injectable silicone: cause of facial nodules, cellulitis, ulceration, and migration. *Aesthetic Plast Surg*. 1996;20(3):267-276.
7. Yedla N, Perez E, Lagari V, Ayala A. Silicone granulomatous inflammation resulting in hypercalcemia: a review of the literature. *AACE Clin Case Rep*. 2019;5(2):e119-e123.
8. Wang LL, Thomas WW, Friedman O. Granuloma formation secondary to silicone injection for soft-tissue augmentation in facial cosmetics: mechanisms and literature review. *Ear Nose Throat J*. 2018;97(1-2):E46-E51.
9. Chen YC, Lee JYY. Extensive disfiguring silicone granuloma of the face successfully treated with corticosteroid and doxycycline. *Int J Dermatol*. 2019;58(11):e217-e219.
10. Broly M, Marie J, Picard C, et al. Management of granulomatous foreign body reaction to fillers with methotrexate. *J Eur Acad Dermatol Venereol*. 2020;34(4):817-820.