Silicone pneumonitis after gluteal filler: a case report and literature review

Boon Hau Ng, Wan Rahiza Wan Mat, Nik Nuratiqah Nik Abeed, Mohamed Faisal Abdul Hamid, Andrea Ban Yu-Lin & Chun Ian Soo

1Pulmonology Unit, Department of Internal Medicine, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia.
2Department of Anesthesiology and Intensive Care Unit, Faculty of Medicine, Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia.

Keywords
Acute respiratory distress syndrome, gluteal filler, silicone embolization syndrome, silicone pneumonitis.

Abstract
Liquid silicone (polydimethylsiloxane) is an inert material that is commonly used for cosmetic purpose. Silicone embolization syndrome (SES) can rapidly progress to pneumonitis as a consequence of the injection of nonmedical-grade liquid silicone. We describe a case of severe silicone pneumonitis complicated with acute respiratory distress syndrome and bilateral pneumothorax secondary to silicone gluteal augmentation. In this case report, we aim to discuss our experience and approach in managing an uncommon case of SES.

Introduction
For decades, Liquid injectable silicone has been used for correction of contour defect or soft-tissue augmentation. Medical-grade liquid silicone (polydimethylsiloxane) becomes the preferred inert material for cosmetic purpose due to its durability, a lack of immunogenicity, and thermal stability; but later found to be associated with silicone embolism syndrome (SES). Liquid injectable silicone induced embolism has been reported by several studies, as a cause of acute pneumonitis with alveolar haemorrhage [1–3].

Case Report
A previously healthy 30-year-old woman presented with three days history of cough with dyspnoea and fever. She had a history of breast augmentation with silicone implant two years ago and had received bilateral gluteal silicone injections from an unlicensed provider one week before the current presentation. The injected volume was approximately 500 mL for each gluteal.

The physical examination was tachycardia, tachypnoea with the respiratory rate of 28 breaths per minute and a temperature of 38°C. Results of the arterial blood gas performed while the patient was breathing room air were as follows: pH, 7.39; PaCO₂, 40 mmHg; PaO₂, 56 mmHg; and peripheral oxygen saturation, 90%. Lungs examination revealed bilateral lower zone crepitations. Other blood investigations which included a complete blood count, comprehensive metabolic panel, and lactate were unremarkable. She was initiated on broad-spectrum antibiotics, oseltamivir, intravenous hydrocortisone 50 mg every 8 h, and 12 L/min (FiO₂: 60%) of high-flow oxygen supplement. However, she developed right-sided pneumothorax and worsened respiratory failure 12 h later.

On chest radiograph, diffuse alveolar opacities in both lung fields were observed. A computed tomographic scan of her thorax demonstrated diffuse bilateral ground-glass infiltrates (Fig. 1). Fiberoptic bronchoscopy was performed, which revealed normal lung segments. Bronchial lung biopsy showed non-refractile lipid
vacuoles, consistent with silicone pneumonitis (Fig. 2). Magnetic resonance imaging of the breast confirmed no intracapsular and extracapsular rupture of the bilateral breast prosthesis. Her bronchial wash was positive for Coronavirus NL63 RNA PCR.

She received lung protective ventilation for acute respiratory distress syndrome (i.e. low tidal volumes and high positive end-expiratory pressure (PEEP)) for continued hypoxia. Unfortunately, two weeks later, she again developed another spontaneous pneumothorax on the left lung and required chest tube drainage. Over four weeks in the intensive care unit, we manage to wean down the ventilatory support. She had an excellent clinical response and discharged with tapering prednisolone over two weeks. At six weeks of outpatient follow up, she was symptom-free, and her chest X-ray revealed residual bilateral reticulations.

Figure 1. A computed tomography scan of the chest (A and B) shows bilateral, diffusely distributed ground-glass opacities with superimposed dependent areas of consolidation. Chest X-ray (C) shows diffuse alveolar opacities of both lung fields. (D) Chest X-ray with bilateral alveolar opacities and pneumothorax with chest tube in-situ. (E) Chest X-ray improvement of bilateral alveolar opacities at 6 weeks of outpatient follow-up.

Figure 2. Trans-bronchial lung biopsy shows lung parenchyma with intra-alveolar haemorrhage, macrophages, and non-refractile vacuole-like structures.
Discussion

The respiratory consequences that associated with silicone injection include acute pneumonitis [4], acute respiratory distress syndrome, alveolar haemorrhages, and pulmonary embolism [5]. The respiratory symptoms generally present within 72 h after injection of higher dose silicone, and a delayed reaction can be seen up to a year and a half after injection [2].

Pulmonary SES results from silicone embolizing to the lungs from an inadvertent intravenous injection or local tissue destruction. The neutrophils ingested the embolization of the silicone material into the lung via the haematogenous or lymphatic and alveolar macrophages induces pneumonitis by local cell-mediated inflammation and release of free radicals and proteolytic enzymes [3]. The release of silicone emboli also leads to occlusion of the microvasculature and trigger the inflammatory response, resulting in pulmonary oedema and haemorrhage.

Diagnosis of the silicone pneumonitis is based on the clinical history of silicone implant or injection, the radiological pattern of subpleural infiltrates and peripherally distributed ground-glass opacities (GGO) [6,7], and tissues biopsy with histopathological features of alveolar haemorrhage [7] and non-refractile vacuole-like structure within the alveoli [1]. Presence of alveolar macrophages with intracytoplasmic silicone inclusions in bronchoalveolar lavage sample is useful in facilitating a diagnosis [3].

Survival is likely, and treatment involves ventilation if necessary and steroids with uncertain utility. The rate of mortality is directly associated with the large volumes of silicone and high-pressure injection [2].

There is no consensus on the treatment of the silicone induced pneumonitis, but case report series show a favourable response with the use of the steroid (Table 1) to reduce the airways inflammation [1,8,9]. General measures include nutrition and ventilation support. Extracorporeal membrane oxygenation (ECMO) may be considered in cases of severe acute respiratory distress syndrome (ARDS) deteriorating on mechanical ventilation. Surgical extraction of the silicone from subcutaneous tissues have had poor outcome due to risk of adverse systemic effects and technically complicated surgery [10].

Carolyn et al. reported a case of acute pneumonitis after silicone injection for gluteal augmentation. The patients presented with haemoptysis, shortness of breath, and acute respiratory failure two days after the silicone injections [11]. Her computed tomography (CT) thorax showed predominantly basilar and peripheral GGO and pulmonary nodules bilaterally. She required ECMO and her condition improved with intravenous methylprednisolone 125 mg every 6 h.

Srilekha Sridhara et al. reported a rare case of silicone pneumonitis with pneumothorax occurred even after 15–20 years of silicone injection [12]. Bronchoscopic lung biopsy demonstrated alveolar interstitium with numerous lipid vacuoles, compatible with silicone deposition. The patient succumbed despite treated with intravenous corticosteroids, broad-spectrum antibiotics, and mechanical ventilation.

Elizabeth et al. reported three cases of breast implants with the onset of symptoms ranges 2–16 years [13]. The CT features of these patients demonstrated diffuse GGO. All three patients responded to prednisolone 40 mg/day.

Our case demonstrates the features of SES after the gluteal silicone filler. No specific site for vascular infiltration was identified in our case; however, many injection marks were observed on the buttocks. MRI breast confirmed that there is no leak from the silicone breast implants and no residual collection in the gluteal region. Thus, the decision not to remove the breast implant and not for surgical exploration of the possibilities of remaining gluteal silicone collection was made after a multidisciplinary meeting comprises of pulmonologist, plastic and reconstructive surgeon, and anaesthetic team. A trans-bronchial lung biopsy was deemed to be a less invasive approach to obtain lung biopsies in order to avoid the risk of general anaesthesia due to poor pulmonary reserve.

The detection of HCoV-NL63 in our patient is incidental. This is consistent with the literature that HCoV-NL63 can be frequently found in asymptomatic individuals [14]. Although, HCoV-NL63 can infects both the upper and lower respiratory tract, it generally associated with mild symptoms, such as fever, cough, pharyngitis, and rhinitis [15]. In rare cases, pneumonia can occur. In total contrast to silicon pneumonitis, lung biopsy histopathological features of HCoV-NL63 pneumonia are chronic pulmonary inflammation, severe alveolar damage, intra-alveolar hyaline membranes, and interstitial oedema [16]. The result of the lung biopsy of our patient was consistent with the diagnosis of silicone pneumonitis.

Learning points for our case includes:

1. Corticosteroids are potentially beneficial in reduces the inflammation of the airways that leading pneumonitis with acute respiratory distress syndrome but does not have any mortality benefits [3,14].
2. Lung-protective ventilation strategies improve survival.
3. Vigilance in the occurrence of pneumothorax in cases of SES especially if complicated with ARDS and requiring mechanical ventilation. Risk of pneumothorax in SES and will usually resolve after chest drain intervention.
4. Trans-bronchial lung biopsy is useful in assisting the diagnosis.
Table 1. Reported cases of pulmonary silicone embolism syndrome.

| Author               | Year | Type of silicone application | Injection site | Symptoms onset after implant | Radiology findings                  | Complications                                      | Treatment                                                                 | Survival |
|----------------------|------|------------------------------|----------------|-----------------------------|-------------------------------------|----------------------------------------------------|---------------------------------------------------------------------------|----------|
| Carolyn et al. [11]  | 2019 | Injection                    | Gluteal        | 2 days                      | Diffuse GGO                         | Pneumonitis, ARDS                               | MTP 125 mg every six hours, ECMO                                        | Yes      |
| Ahad et al. [17]     | 2019 | Implant                      | Breast         | 14 years                    | Alveolar infiltrates                | Pneumonitis                                      | Corticosteroids, ECMO, antibiotics                                      | No       |
| Srilekha et al. [12] | 2018 | Injection                    | —              | 15–20 years                 | Diffuse GGO                         | Pneumonitis, pneumothorax                        | Corticosteroids                                                          | No       |
| Elizabeth et al. [13]| 2018 | Implant                      | Breast         | 16 years                    | Hilar & mediastinal LN, pulmonary nodules | Pulmonary nodules                           | Prednisone 40 mg/day tapering down over 6 months                      | Yes      |
|                     |      | Implant                      | Breast         | 2 years                     | Diffuse micro-nodules with GGO      | Pneumonitis                                       | Implant removal, prednisone 40 mg/day                                 | Yes      |
|                     |      | Implant                      | Breast         | 12 years                    | Diffuse GGO and reticular opacities | Organising pneumonia                           | Prednisone 40 mg/day with tapering down over 6 months and azathioprine as steroid sparing agent | Yes      |
| Arthur et al. [18]   | 2018 | Implant                      | Breast         | 6 months                    | Alveolar infiltrates, diffuse GGO   | Pneumonitis, PH                                 | Implant removal, prednisolone 40 mg/day, ECMO                           | Yes      |
| Rafael et al. [19]   | 2017 | Implant                      | Gluteal        | 4 months                    | —                                   | Granulomatous inguinal LN                       | Capsulotomy                                                              | Yes      |
| María et al. [20]    | 2016 | Implant                      | Breast         | 10 years                    | Consolidation                       | Pneumonitis                                      | Implant removal, corticosteroids                                      | Yes      |
| Kirill et al. [21]   | 2016 | Injection                    | Gluteal        | 2 months                    | Diffuse GGO                         | Pneumonitis                                      | Supportive                                                                | Yes      |
|                     |      | Injection                    | Gluteal        | 8 months                    | Diffuse GGO, mediastinal & hilar LN | Pneumonitis                                      | Supportive                                                                | Yes      |
| Ayush et al. [22]    | 2016 | Implant                      | Breast         | 18 years                    | Diffuse GGO                         | Pneumonitis                                      | Implant removal, corticosteroids                                      | Yes      |
| Erin et al. [23]     | 2015 | Injection                    | Gluteal        | 5 months                    | Alveolar infiltrates                | Gluteal abscess, ARDS                            | Incision and drainage, antibiotics                                      | Yes      |
| Alex et al. [24]     | 2013 | Injection                    | Gluteal        | 2 days                      | Diffuse GGO                         | Pneumonitis, PH                                 | MTP 125 mg every six hours                                           | Yes      |
| Dercio et al. [25]   | 2012 | Injection                    | Gluteal        | 2 weeks                     | Alveolar infiltrates                | Pneumonitis                                      | Corticosteroids                                                          | Yes      |
| Author          | Year | Type of silicone application | Injection site          | Symptoms onset after implant | Radiology findings                              | Complications                  | Treatment          | Survival |
|-----------------|------|------------------------------|--------------------------|------------------------------|-----------------------------------------------|-------------------------------|-------------------|----------|
| Denyo et al. [26] | 2012 | Injection                    |                          | 36–48 h                      | Alveolar infiltrates                         | Pneumonitis, PH              | —                 | —        |
|                 |      | Injection                    |                          |                              |                                               | Pneumonitis, PH              | —                 | —        |
| Priya et al. [27] | 2011 | Injection                    | Gluteal                  | 6 h                          | Diffuse GGO                                   | Pneumonitis, PH              | Supportive        | Yes      |
| Sophie et al. [28] | 2010 | Injection                    | Gluteal, hip             | 1 day                        | Noncalcified pulmonary nodules, GGO           | Pneumonitis                   | MTP               | Yes      |
| Rupen et al. [29]  | 2008 | Injection                    | Thigh                    | 3 days                       | Alveolar infiltrates                         | Pneumonitis                   | MTP               | Yes      |
| Richard et al. [30] | 2008 | Injection                    | Gluteal, thigh, face     | 4 h                          | Alveolar infiltrates                         | Organising pneumonia, ARDS   | MTP               | No       |
| Rafael et al. [31]  | 2007 | Injection                    | Breast                   | 40 h                         | Alveolar infiltrates                         | Pneumonitis, ARDS            | Supportive        | No       |
| Grigoriy et al. [32] | 2006 | Injection                    | Gluteal                  | 12 days                      | Alveolar infiltrates                         | Pneumonitis, PH              | MTP 250 mg every six hours | Yes      |
| Samuel et al. [33]  | 2006 | Injection                    | Gluteal                  | —                            | Subpleural GGO and consolidation             | Pneumonitis, PH              | Supportive        | -        |
| Alex et al. [34]  | 2004 | Injection                    | Breast                   | 1 week                       | Alveolar infiltrates                         | Pneumonitis, PH              | Corticosteroids    | Yes      |
| Cheol et al. [35]  | 2003 | Injection                    | Vaginal colpoplasty      | 2 days                       | Interstitial infiltrates, airspace consolidation | Pneumonitis                   | Corticosteroids    | Yes      |
| Jean et al. [3]   | 1983 | Injection                    | Trochanter               | 3 days                       | —                                             | Pneumonitis                   | Supportive        | Yes      |
|                   |      | Injection                    | Trochanter               | 2 days                       | —                                             | Pneumonitis                   | Supportive        | Yes      |
|                   |      | Injection                    | Trochanter               | 1 day                        | Interstitial infiltrates, airspace consolidation | Pneumonitis                   | Supportive        | Yes      |

ARDS, acute respiratory distress syndrome; ECMO, extracorporeal membrane oxygenation; GGO, ground glass opacities; LN, lymphadenopathy; MTP, methylprednisolone; PH, pulmonary haemorrhages.
5. Patients should be advised that there is a risk that silicone injections can be associated with serious pulmonary complications.

The injection of silicone for cosmetic purpose is debatable due to the sequelae of the pneumonitis and the potential for pulmonary toxicity in asymptomatic patients who receive silicone injections. Bronchoalveolar lavage and trans-bronchial lung biopsy can help to confirm the diagnosis. This case report serves to highlight an emergent danger associated with illicit silicone use for cosmetic purposes and clinicians should be aware of the potential complications.

Disclosure statement
Appropriate written informed consent was obtained for publication of this case report and accompanying images.

References
1. Chung KY, Kim SH, Kwon IH, et al. 2002. Clinicopathologic review of pulmonary silicone embolism with special emphasis on the resultant histologic diversity in the lung—a review of five cases. Yonsei Med. J. 43(2):152–159.
2. Schmid A, Tzur A, Leshko L, et al. 2005. Silicone embolism syndrome: a case report, review of the literature, and comparison with fat embolism syndrome. Chest 127(6):2276–2281.
3. Chastre J, Basset F, Viau F, et al. 1983. Acute pneumonitis after subcutaneous injections of silicone in transsexual men. N. Engl. J. Med. 308(13):764–767.
4. Parikh R, Karim K, Parikh N, et al. 2008. Case report and literature review: acute pneumonitis and alveolar hemorrhage after subcutaneous injection of liquid silicone. Ann. Clin. Lab. Sci. 38(4):380–385.
5. McCurdy HH, and Solomons ET. 1977. Forensic examination of toxicologic specimens for dimethylpolysiloxane (silicone oil). J. Anal. Toxicol. 1(5):221–223.
6. Restrepo CS, Artunduaga M, Carrillo JA, et al. 2009. Silicone pulmonary embolism: report of 10 cases and review of the literature. J. Comput. Assist. Tomogr. 33(2):233–237.
7. Price EA, Schueler H, and Perper JA. 2006. Massive systemic silicone embolism: a case report and review of literature. Am. J. Forensic Med. Pathol. 27(2):97–102.
8. Zamora AC, Collard HR, Barrera L, et al. 2009. Silicone injection causing acute pneumonitis: a case series. Lung 187(4):241–244.
9. Clark RF, Cantrell FL, Pacal A, et al. 2008. Subcutaneous silicone injection leading to multi-system organ failure. Clin. Toxicol. 46(9):834–837.
10. Sanz-Herrero F, de Casimiro-Calabuig E, and Lopez-Miguel P. 2006. Acute pneumonitis after subcutaneous injection of liquid silicone as a breast implant in a male-to-female transsexual. Arch. Bronconeumol. 42(4):205–206.
11. Carolyn Frances M, and Zachary Paul R. 2019. Pulmonary embolism secondary to silicone injection. Int. J. Med. Stud. 7(2):41–44.
12. Srilekha Sridhara MB, Batool K, Chowdhury J, et al. 2018. A rare case of delayed chronic pneumonitis following non-medical grade silicone injections in a transgender woman. J. Unexplored Med. Data 3(1):1–5.
13. Elizabeth F, Dror R, Eina E, et al. 2018. Interstitial lung diseases associated with metal content in silicone breast implants: a case series. Sarcoidosis Vasc. Diffuse Lung Dis. 35(4):381–389.
14. van der Zalm MM, van Ewijk BE, Wilbrink B, et al. 2009. Respiratory pathogens in children with and without respiratory symptoms. J. Pediatr. 154(3):396–400.
15. Bastien N, Anderson K, Hart L, et al. 2005. Human coronavirus NL63 infection in Canada. J Infect Dis 191(4):503–506.
16. Oosterhof L, Christensen CB, and Sengelov H. 2010. Fatal lower respiratory tract disease with human corona virus NL63 in an adult haematopoietic cell transplant recipient. Bone Marrow Transplant. 45(6):1115–1116.
17. Azeeem A, Khuwaja S, Parthvi R, et al. 2019. Pulmonary fibrosis and embolism secondary to silicone implant leak. BMJ Case Rep 12:e229470.
18. Ng AF, and Elmann EM. 2018. VV ECMO rescue of silicone syndrome. Ann Clin Case Rep 3:1546.
19. Biguria R, and Ziegler OR. 2017. Silicone migration after buttock augmentation. Plast Reconstr Surg Glob Open 5(12):e1583.
20. Hernández MJG, Milena GL, and Carazo ER. 2016. Subacute silicone pneumonitis after silent rupture of breast implant. Arch Bronconeumol 52(7):393–400.
21. Lyapichev K, Chinea FM, Poveda J, et al. 2016. Pulmonary empty spaces: silicone embolism—a decade of increased incidence and its histological diagnosis. Case Rep Pathol 2016:3741291.
22. Ayush A, Inaty H, Mukhopadhyay S, et al. 2016. Chronic pulmonary silicone embolism related to saline breast implants. Ann Am Thorac Soc 13(1):139–141.
23. Purdy-Payne EK, Green J, Zenoni S, et al. 2015. A serious complication of illicit silicone injections: latent silicone embolization syndrome after incision and drainage of local injection site. Surg Infect 16:473–477.
24. Essénmacher AC, and Astani SA. 2013. Respiratory disease following illicit injection of silicone: a case report. Case Rep Med 2013:743842.
25. Mendonca D, Leitao DS, Friend R, et al. 2012. An unusual case of pulmonary embolism. Respir Care 57(8):1345–1347.
26. Zakhia DA, Bertucci C, Alasii O, et al. 2012. Subcutaneous silicone injection and the silicone embolism syndrome: a clinicopathologic correlation of two cases. Am J Clin Pathol 138:A287.
27. Gopie P, Sakhamuri S, Sharma A, et al. 2011. Acute pneumonitis secondary to subcutaneous silicone injection. Int J Gen Med 4:477–479.

28. Bartsich S, and Wu JK. 2010. Silicone emboli syndrome: a sequela of clandestine liquid silicone injections. A case report and review of the literature. J Plast Reconstr Aesthet Surg 63:e1ee3.

29. Parikh R, Karim K, Parikh N, et al. 2008. Acute pneumonitis and alveolar hemorrhage after subcutaneous injection of liquid silicone. Ann Clin Lab Sci 38(4):380–385.

30. Richard F, Clark F, Cantrell L, Pacal A, et al. Subcutaneous silicone injection leading to multisystem organ failure. Clin Toxicol 46(9):834–837.

31. de March Ronsoni R, Schwingel FL, Melo LH, et al. 2007. Pulmonary embolism due to liquid silicone: case report. Respir Med Extra 3:172–174.

32. Gurvits GE. 2006. Silicone pneumonitis after a cosmetic augmentation procedure. N Engl J Med 354:211–212.

33. Dawn SK, Elicker BM, Leung JWT, et al. 2006. The silicone syndrome. Clin Pulm Med 13:146–147.

34. Rosioreanu A, Brusca-Augello GT, Ahmed QAA, et al. 2004. CT visualization of silicone-related pneumonitis in a transsexual man. Am J Roentgenol 183:248–249.

35. Cheol K, Doo C, Chul-Gyu Y, et al. A case of acute pneumonitis induced by injection of silicone for coloplasty. Respiration 70:104–106.