Siliconosis: an unknown entity in aesthetic breast surgery

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ABSTRACT

Background: Siliconosis was first described in the late 1980s and it is still remaining as a rare condition and mainly as a diagnosis of exclusion after breast cancer investigation. There are only limited cases reported and published with even smaller amount related to the breast implants (31 studies and 8 related to breast implants found in PubMed search in December 2019).

Case presentation: Authors present the case of siliconosis secondary to bilateral cosmetic breast augmentation performed back in 1989. The patient was troubled with a myriad of complications and underwent an exchange of implants with Trilucent™ implants in 1997 and their subsequent removal in 1998. Later, they underwent bilateral mastopexy and is currently free of implants. Ever-since the first operation patient complained of pain, localized tenderness, swelling, axillary fullness, paraesthesia and partial paralysis in her upper limbs amongst other symptoms. Objective investigations including plain radiographs, USS, CT, MRI, nerve conduction studies, rheumatological screen yielded essentially negative results. Thoroughly investigation for breast cancer was conducted, including several operations and biopsies of axillary swellings with confirming reactive lymphadenopathy. A working diagnosis of siliconosis has been made and the patient was treated expectantly.

Conclusion: This case should remind our colleagues of the ethical and professional responsibilities we have toward our patients in explaining all the potential risks involved in breast augmentation and also to keep an open mind when meeting patients complaining of systemic symptoms post breast augmentation.

Keywords: Syliconosis, autoimmune/inflammatory syndrome induced by adjuvants (ASIA), silicone implant incompatibility syndrome (SIIS), breast augmentation, breast implants.

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Background

Siliconosis was described few decades ago. Nevertheless, it remains difficult to diagnose. Patients often need lengthy investigations putting up with the vague, non-specific symptoms which markedly impact quality of life. There are limited data relating the condition in the literature, thus authors believe this case will be useful addition.

Case Presentation

A 56-year-old patient was first referred to our care in 1998 after suffering with mastalgia and axillary discomfort which were attributed to her bilateral breast augmentation in the late 1980s. Patient first underwent bilateral breast augmentation with silicone implants though submammary fold access in 1989. Following this, in March 1997, they developed left sided mastalgia and left axillary lump. This was investigated with ultrasound and excision biopsy which showed reactive lymphadenitis secondary to silicone exposure. An MRI of both breasts and axillae showed a rupture of the left implant whilst the right implant remained intact. Due to the rupture, the patient opted for removal of the silicone implants and their replacement with Trilucent™ implants.

Following a minor wound infection and a 12 month's symptom free period, patient presented in the clinic with ridging of the skin and a cystic mass in the right breast upper outer quadrant. Due to a positive family history (maternal aunt) for the breast cancer, they were reviewed in breast clinic. A benign breast nodule was diagnosed. Whilst breast ultrasound showed no abnormality, patient continued to suffer from left sided breast mastalgia and complained of intermittent axillary swelling. Later that year, in December 1998, the removal of both Trilucent™ implants and bilateral superior pedicle mastopexy were conducted as a patient's choice. Findings at the time of the operation showed normal right implant and capsule. The left implant was intact, however surrounded by odorless milky fluid and capsular calcium deposits. Capsular fluid sample tests depicted raised triglyceride levels with no evidence of an infective process.

In 1999, patient returned to our care with bilateral axillary lymphadenopathy and worsening mastalgia. She had developed anxiety as there were growing concerns related to Trilucent™ implants which had been investigated by Medical and Healthcare Products Regulatory Agency (MHRA). In March 2000, the MHRA officially issued a

warning for the removal of Trilucent™ implants due to the risk of production of genotoxic products from the breakdown of the soya bean oil filler [10].

In the following year, patient continued to suffer from generalized weakness and mastalgia in the left breast. The mastopexy and lymph node biopsies both had confirmed reactive lymphadenitis, whilst all her blood tests were normal. Patient developed new paraesthesia like symptoms and pain deep to her chest and down left arm in the following years. They were fully investigated by rheumatology and neurology for this neurogenic pain and autoimmune disorders. These investigations did not find any cause of her symptoms. Both ultrasound and computerized tomography did not reveal any abnormality.

Due to the debilitating nature of their symptoms' patient was offered a mastectomy and free flap reconstruction. They were reviewed by neuropsychology and jointly decided it would be in her best interests. Further biopsies of lymph node showed foreign body reaction.

At this stage tissue biopsies have been sent to specialized distant laboratory for specific multiple displacement amplification DNA adducts in tissues which have been associated with Trilucent™ implants. After exhaustive investigation, there were no oil filler products identified while histopathology tests performed on the biopsies did not show any silicone gel or refractile particles.

Patient continued being symptomatic with increasing of non-specific symptoms despite all tests performed in the UK were negative. In May 2005, MHRA decided to forward histology slide to the Trilucent™ specialist panel in the USA. After reviewing her case notes and multiple biopsies slides, the Vanderbilt University Medical Centre issued a report summarizing that the histology slides from 1998 show characteristic reactive changes associated with fibrous capsule around the implant. Some material compatible with silicone shell and abundant foreign body reaction to lipid like material near the capsule. This extended to a small fragment of skeletal muscle. A report issued by the New York Medical College Laboratory supported that the breast pathology was unlikely to be related to the Trilucent™ implants but reinforced the suspicion that this was related to her original silicone implants.

Furthermore, patient developed a non-specific allergic reaction to hair-dye and was diagnosed with allergic tendency following high IgE levels.

All reports and results were fully disclosed to the patient who has since been treated symptomatically and expectantly on the basis of a siliconosis diagnosis.

Discussion

Siliconosis was first described in the late 1980s under the generic term "the adjuvant breast disease". In 2011, Shoenfeld [11] described a group of disorders: siliconosis, the Gulf war syndrome, the macrophagic-myofasciitis syndrome and post-vaccination phenomena which share similar signs and symptoms under a term ASIA induced by adjuvants. Later, Silicone Implant Incompatibility syndrome (SIIS) was recognized as a subtype of ASIA [12] (The suggested criteria for ASIA syndrome as adopted by Shoenfeld et al., are seen in Figure 1)

Even though systemic effects remain controversial, there are many well-known localized complications to silicone breast implants including implant rupture, silicone leakage through shell, capsular fibrosis, and contracture. The proposed mechanism of action is the induction of autosilicone antibodies promoting an autoimmune reaction [6]. Silicone breast implants have been also recognized as inducers of a chronic inflammatory response when the silicone migrates, likened to an autoimmune rheumatic disease and fibromyalgia [3,7].

Nevertheless, siliconosis is remaining a rare condition and remain mainly as a diagnosis of exclusion after breast cancer investigation with even smaller amount related to the breast implants (31 studies and 8 related to breast implants found in PubMed search in December 2019).

Silicone was initially considered as an inert substance. This was soon disproved with the presentation of many patients undergoing silicone breast implants who reported a variety of symptoms which did not fit any connective tissue disorder diagnosis. Brown et al. [1] conducted a large cohort study on 907 patients who underwent silicone implantation by contacting a questionnaire with questions on health status, implant satisfaction, symptoms of connective tissue diseases, and known diagnosis of connective tissue disease. These patients then went on to have an MRI to assess the implant status. Their results showed there was an increase odds ratio in patients developing fibromyalgia after the rupture of silicone breast implants (odd ratio 2.8, 95% CI 1.2-6.3) and connective tissue disorders (odd ratio 2.6, 95% CI 0.8-8.5). A meta-analysis published the year before in 2000 by Janowsky et al. [8] did not show an association and the risk of developing a connective tissue disorder following silicone breast implantation was 0.80 (95% CI 0.62-1.04). However, there have been numerous studies and case series suggesting an association [2,9].

The recent large study by Colaris et al. [4] compared a historical cohort of 100 women from 1985 to 1992 with adjuvant breast disease to a cohort of 100 women with SIIS diagnosed in 2014. They found that the symptom profile in these two groups was comparable and that improvement in symptoms was often noted post explanation in around 50% of women. By contrast, multiple case reports analyzed in the most recent literature review shown no major improvement of the symptoms after removal of implants as it happened in our case.

Fuzzar et al. [12] proposed an algorithm for treatment of silicone implant incompatibility syndrome incorporating conclusions of recent studies [5,13] and based on the existing studies over the course of 30 years included in literature review.

Major Criteria:

- Exposure to external stimuli (infection, vaccine, silicone, adjuvant) prior to clinical presentation
- The presence of 'typical' clinical manifestations:
  - Myalgia, myositis or muscle weakness
  - Arthralgia and/or arthritis
  - Chronic fatigue, un-refreshing sleep or sleep disturbance
  - Neurological manifestations (especially associated with demyelination)
  - Cognitive impairment, memory loss
  - Pyrexia and dry mouth
- Removal of inciting agent induces improvement
- Typical biopsy of involved organs

Minor Criteria:

- The appearance of autoantibodies directed at the suspected adjuvant
- Other clinical manifestations (i.e. irritable bowel syndrome)
- Specific HLA (i.e. HLA DRB1, HLA DQB1)
- Evolvement of an autoimmune disorder (i.e. multiple sclerosis, sarcoidosis)

Figure 1. *Autoimmune/inflammatory syndrome induced by adjuvants criteria.*

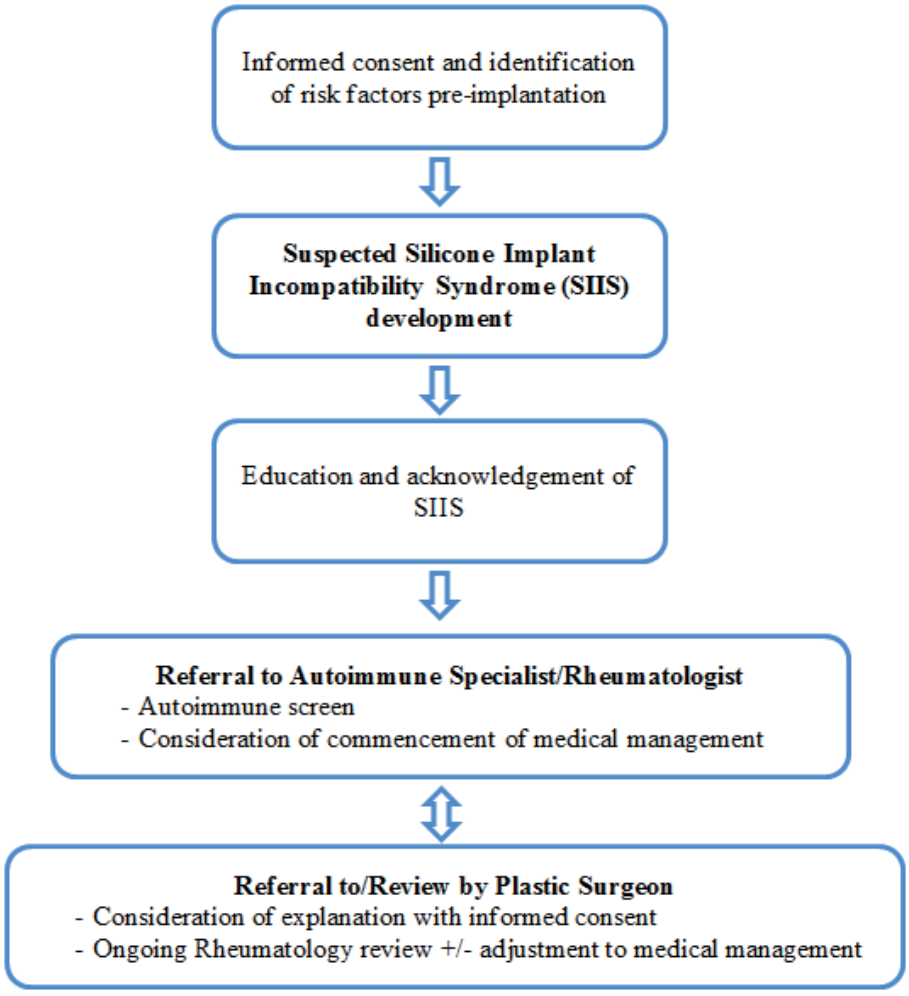

Figure 2. *Algorithm for the treatment of silicone implant incompatibility syndrome.*

Conclusion

Case we present is a puzzling one with several factors including two different types of breast implants challenging the diagnosis, consistency in treating the patient and seeking help from other colleagues, the diagnosis of siliconosis at the patient fits most of the major criteria of ASIA.

We present this case to remind our colleagues of the ethical and professional responsibilities we have towards our patients in explaining all the potential risks involving in breast augmentation and to remind them to keep an open mind when meeting patients complaining of systemic symptoms post breast augmentation. This point is well supported by the algorithm mentioned earlier with the informed consent and identification of risk factors put on top of it.

With the increasing frequency of breast augmentation surgeries involving silicone implants, the risk of implant rupture, silicone migration with the long term inflammatory conditions associated with siliconosis should not be disregarded. Presenting symptoms including myalgias, arthralgias, chronic fatigue, sleep disturbance and cognitive impairment are vague and largely subjective but must be taken seriously in the patient with breast implants. Potential alternative diagnoses should be looked at in multidisciplinary team including medical doctors, rheumatologist, immunologist or other health care professional can be of great assistance in outlining a diagnostic and treatment plans.

What is new?

This diagnosis is a grey area in literature especially in relation to the patient diagnosis. Often a diagnosis of siliconosis is made as a diagnosis of exclusion when all other options came back as negative.

List of Abbreviations

ASIA	Autoimmune syndrome induced by adjuvants
MHRA	Medical and Healthcare Products Regulatory Agency
SIIS	Silicone implant incompatibility syndrome

Consent for publication

Written informed consent was taken from the patient.

Ethical approval

Ethical approval is not required at our institution for publishing an anonymous case report.

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Summary of the case

1	**Patient (gender, age)**	56, Female
2	**Final diagnosis**	Siliconosis
3	**Symptoms**	Mastalgia and axillary discomfort
4	**Medications**	Treatment with surgical procedurs
5	**Clinical procedure**	CT, MRI, Immuno-histological investigations
6	**Specialty**	Plastic Surgery