

Thoracic Outlet Syndrome Following Breast Implant Rupture

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Summary: We present a patient with bilateral breast implant rupture who developed severe locoregional silicone granulomatous lymphadenopathy. Poly Implant Prothese silicone implants had been used for bilateral breast augmentation 5 years prior. Extracapsular implant rupture and bilateral axillary lymphadenopathy indicated explantation, capsulectomy, and selective lymph node excision. Histology demonstrated silicone lymphadenopathy with no evidence of malignancy. Over the subsequent 12 months, she developed progressive locoregional lymphadenopathy involving bilateral cervical, axillary, and internal mammary groups, resulting in bilateral thoracic outlet syndrome. We report the unusual presentation, progression, and the ultimate surgical management of this patient. (*Plast Reconstr Surg Glob Open* 2015;3:e331; doi: 10.1097/GOX.0000000000000295; Published online 18 March 2015.)

CASE REPORT

A 33-year-old woman presented with bilateral breast pain 5 years following submuscular augmentation mammoplasty. There was no history of trauma. The implants were textured silicone Poly Implant Prothese (PIP) 390 cm³ implants. Examination revealed bilateral tender D-cup breasts and tender axillary lymph nodes. Magnetic resonance imaging (MRI) demonstrated bilateral extracapsular implant rupture with bilateral internal mammary and axillary lymphadenopathy (Fig. 1). The patient elected against having the implants replaced. She underwent explantation and total capsulectomy. A selective excision of palpably enlarged bilateral level 1 axillary lymph nodes was also performed at her request. His-

tology demonstrated a fibrous pseudocapsule with silicone material throughout and silicone lymphadenopathy with no evidence of malignancy. The lymph nodes showed partial effacement of the architecture by nonnecrotizing granulomatous inflammation, with a prominent foreign body giant cell reaction. Clear vacuoles were seen within the cytoplasm, consistent with the presence of silicone (Fig. 2).

Four months following surgery, she presented with a tender left supraclavicular mass. She also complained of intermittent glove-like paresthesiae of her left hand; however, neurological examination was normal. MRI demonstrated bilateral lymphadenopathy of the axillae and supraclavicular regions and a single enlarged mediastinal node. Following discussion with a hematologist with a specialist interest in lymphoproliferative disease, she underwent excision biopsy of 5 left-sided supraclavicular nodes to exclude lymphomatous malignancy. Histology revealed silicone lymphadenopathy with no evidence of malignancy. Immunophenotyping of the nodes revealed a polyclonal B-cell population, and peripheral bloods showed an increased concentration of natural killer cells. A bone marrow biopsy was performed to further investigate the natural killer cell

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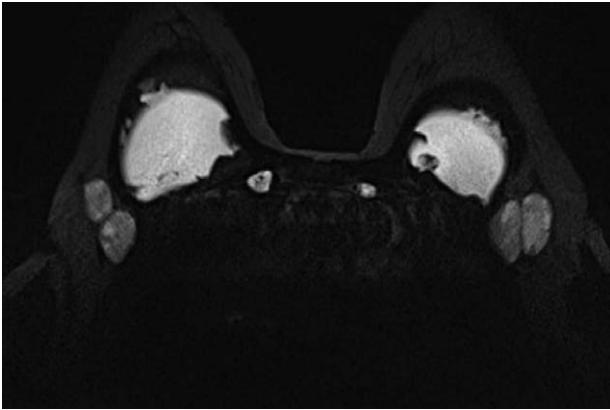


Fig. 1. Axial MRI from first presentation demonstrating extracapsular implant rupture and axillary and internal mammary lymphadenopathy.

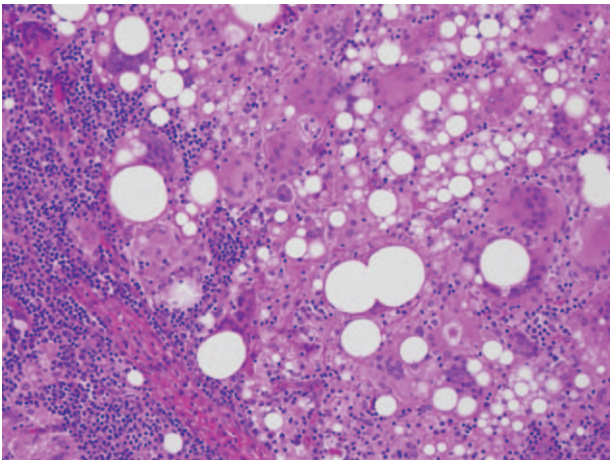


Fig. 2. Histology slide of lymph node demonstrating silicone lymphadenopathy, H&E $\times 100$.

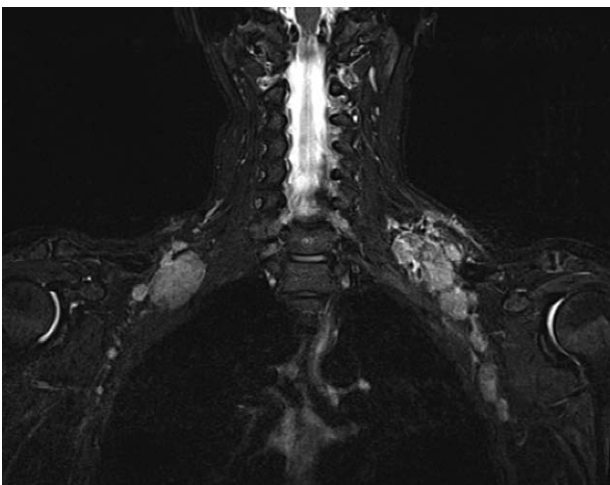


Fig. 3. Coronal MRI demonstrating mass effect on bilateral brachial plexuses.

population and demonstrated no evidence of lymphoproliferative disease. A trial of prednisolone was commenced to suppress the granulomatous reaction. Clinically the lymphadenopathy and paresthesia subsided within 2 weeks of commencing steroids. After 4 weeks, however the steroids were ceased due to extensive folliculitis.

She re-presented 4 months later with a tender left supraclavicular mass. MRI demonstrated bilateral axillary, internal mammary, supraclavicular, and upper paratracheal lymphadenopathy. This extended along the course of the brachial plexuses with mild mass effect. In order to exclude lymphoproliferative change, she underwent excision biopsy of 3 left-sided supraclavicular nodes and 1 right-sided supraclavicular node. Histology revealed silicone lymphadenopathy with no evidence of malignancy.

Three weeks following surgery, she presented with tender bilateral supraclavicular swelling, coldness, and numbness in both arms and hands, which felt “dead” on waking but were relieved by shaking. Examination revealed altered sensation and reduced power in C4-T2 distribution and positive Adson’s and Roos’ provocative tests for thoracic outlet compression. MRI images were unchanged. The majority of the mass effect was exerted at the level of the brachial plexus divisions and cords (Fig. 3). Given the severity and progressive nature of her symptoms, along with the radiological findings and after discussion with the hematologist, we recommended she undergo targeted excision of enlarged lymph nodes in the proximity of the brachial plexus. This was performed via supraclavicular to deltopectoral incisions and axillary incisions (Fig. 4). On the right, the clavicle was split for access and later osteosynthesised (Fig. 5). Bilateral brachial plexuses were exposed at level V and enlarged nodes excised. In addition, a bulky level IV node was excised on the right, and bulky level III/IV nodes were excised on the left. Enlarged nodes from bilateral axillary level II and



Fig. 4. Preoperative photograph of the proposed supraclavicular to deltopectoral incisions used to access the bilateral brachial plexuses. Previous biopsy incisions were incorporated. The acromions are also marked.

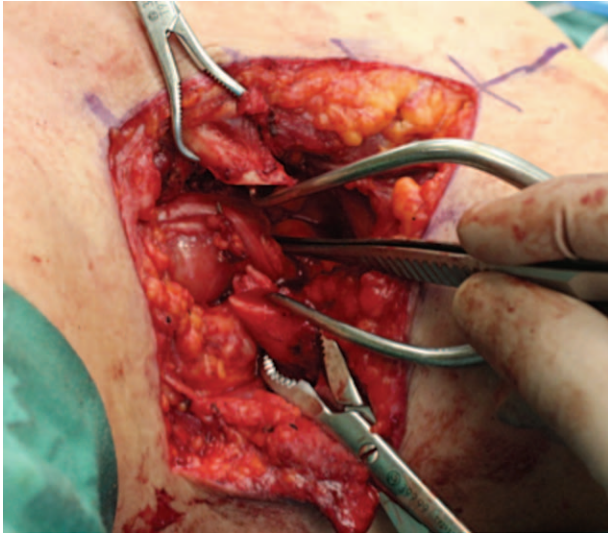


Fig. 5. Intraoperative photograph of the root of the right neck taken from a cephalic viewpoint. The clavicle is divided, and the displacement effect of anterior and posterior enlarged lymph nodes on the brachial plexus can be seen.

III were excised. Histology revealed silicone lymphadenopathy with no evidence of malignancy. Within a day of surgery, her hand sensation improved and provocative tests were negative. Over the following 3 months, she regained normal sensation, power, and range of motion.

Six months following surgery, she presented with symptoms of thoracic outlet compression on the left side. Examination revealed tender swelling in the left supraclavicular fossa, altered sensation and reduced power in C4-T2 distribution, and positive Adson's and Roos' test. MRI demonstrated asymmetrically enlarged left supraclavicular lymph nodes closely associated with the cords of the brachial plexus. Following further multidisciplinary discussion, she underwent an en bloc level 5 lymph node clearance. A significant number of enlarged nodes were cleared under omohyoid, trapezius, and inferior to the clavicle. Histology revealed silicone lymphadenopathy with no evidence of malignancy. Four months following surgery, she regained normal sensation, power, and range of motion, with no evidence of lymphoedema.

DISCUSSION

This is the first documented case of silicone lymphadenopathy resulting in thoracic outlet syndrome. The patient presented 4 times over the course of 12 months with lymphadenopathy. Given the differential diagnosis of lymphoma, a hematologist was consulted. Analysis of peripheral blood, bone marrow biopsy, and lymph node biopsy was performed at each stage to exclude lymphoproliferative change.

In this case, the silicone appeared to stimulate a chronic granulomatous reaction. Despite its initial reputation as a biologically inert substance, silicone has been associated with local and systemic granulomatous inflammatory reactions affecting breast tissue and lymph nodes. When leakage occurs, silicone can be transported to regional lymph nodes by macrophages in the reticuloendothelial system resulting in chronic foreign body granulomatous reactions. In the majority of cases, silicone migration is limited to the axillary lymph nodes. However, spread to other groups is possible.¹⁻⁴

A recent worldwide review of patients with implant-associated anaplastic large cell lymphoma (ALCL) showed that 17% of patients presented with axillary lymphadenopathy.⁵ As such, it was important to exclude implant-associated ALCL. In general, implant-associated ALCL has an indolent clinical course with the mean interval between implant insertion and lymphoma diagnosis being 9 years (range, 1–32 years).⁵ To date, no cases of ALCL have been diagnosed in patients with PIP implants. The risk of malignant transformation is not known for PIP implants or silicone granulomatous disease, and therefore, we plan to monitor this patient's progress long term from both hematological and surgical point of view.

The breast implants in this case were manufactured by PIP. This is noteworthy to mention as the sale of these implants was suspended in March 2010 after the French Health Products Safety Agency found that the silicone gel used by PIP was not medical grade and therefore did not comply with Conformité Européenne marking.⁶ Physicochemical analysis has demonstrated that PIP implants have increased levels of low-molecular-weight siloxanes compared with medical-grade breast implants. However, cytotoxic and genotoxic tests have shown no acute toxic effect. Clinically, these implants have been shown to have a higher rupture rate than medical-grade implants.^{7,8} Cases of PIP implant rupture associated silicone lymphadenopathy have been reported in the literature. In all of these cases, the silicone has migrated to cervical and/or axillary lymph nodes.^{1,4,8-10}

There is no current consensus on the management of silicone granulomatous disease.^{2,4} Options include conservative, immunosuppression, or surgical excision. Recurrence has been reported following surgical excision.² There is a paucity of literature on immunosuppressive therapy for silicone granulomatous disease. In this case, the patient did not tolerate corticosteroid therapy, and therefore, it was ceased. Alternative immunosuppressants were not used due to their cytotoxic side effects. She then developed further lymphadenopathy over a period of 12 months, with worsening neuropathic symptoms.

However, as the symptoms were becoming more severe, with MRI confirmation of mass effect, we proposed a more comprehensive excision of grossly enlarged lymph nodes for symptom relief. Given the necessary cutaneous scars for access and the potential risks of such an operation, these decisions were made after careful consideration and discussion with orthopedic, thoracic, and vascular surgeons, the hematologist, and the patient. Complete cervical and axillary lymph node clearance was not planned at this stage due to the risk of lymphoedema. Although initially effective in symptom relief, when she presented 6 months later with left-sided thoracic outlet symptoms, the decision was made to formally clear the level 5 lymph nodes encroaching on the brachial plexus. This has again resolved her symptoms, accepting that lymphoedema may still present as a long-term complication. We propose indications for surgical excision include biopsy to exclude malignancy and symptomatic relief where medical management has failed.

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PATIENT CONSENT

Patient provided written consent for the use of her images.

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