

pleura are small, many are huge pedunculated tumors, as was the case in the younger patient reported on here. The malignant tumors are often > 10 cm on presentation.⁵ The tumors are present for many years and do not cause symptoms until they are quite large. While the daughter's larger tumor was found because of local symptoms, the mother's smaller tumor was detected incidentally on imaging of the chest. Although not present in our two patients, benign SFTs of the pleura may be associated with paraneoplastic syndromes, especially clubbing and hypoglycemia.

A review of the English-language literature failed to reveal any other documented occurrence of familial SFTs, at any anatomic site. A MEDLINE search of "solitary fibrous tumor" combined with "familial" yielded a report of two siblings with calcifying fibrous tumors.⁶ However, the tumors described in that report are a different entity. They are CD-34 nonreactive, and they contain psammomatous or dystrophic calcification, a feature that is not found in SFTs.

The presence of SFTs of the pleura in a mother and daughter may be due to chance, exposure to a common environmental agent, or due to a germ-line mutation that was genetically transmitted. An SFT of the pleura is an uncommon neoplasm. Only about 800 cases have been reported.⁷ The gene or chromosome responsible for SFTs of the pleura has not yet been identified. Different cytogenetic studies of SFTs of the pleura have reported^{8–11} varying mutations in chromosome number (eg, trisomy 8, trisomy 21, and trisomy 5). Miettinen and coworkers,¹⁰ using comparative genomic hybridization, showed that changes in DNA copy number occurred more commonly in those SFTs that were > 10 cm and in those with greater mitotic activity. Various more complex translocations have been found as well. A break point in chromosome 4 was found in case reports of SFTs in both pleural and peritoneal locations,^{12,13} but this has not been confirmed in other studies. Given that no consistent chromosomal abnormality has been reported, further cytogenetic or molecular genetic investigations of SFTs of the pleura are needed before their genetic etiology can be known.

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Silicone Thorax Due to a Ruptured Breast Implant*

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A woman with a history of bilateral mastectomy and silicone implants for fibrocystic disease and a history of atrial septal defect repair presented with pleural nodules on a chest radiograph. A thorascopic biopsy performed for possible mesothelioma demonstrated chronic inflammation and focal pleural fibrosis due to a foreign-body reaction secondary to silicone. This was confirmed using scanning electron microscopy and energy-dispersive radiograph elemental analysis. As the population ages, the increasing frequency of ruptured silicone implants and the need for heart surgery may result in a corresponding increase in the risk for fibrothorax secondary to inadvertent silicone introduction during surgery.

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Key words: cardiac surgery; fibrothorax; silicone breast implants; silicone thorax

Abbreviations: EDXEA = energy dispersive X-ray elemental analysis; EM = scanning electron microscopy

More than 1.5 million women in the United States have silicone breast implants.¹ Due to safety concerns, in 1992 the US Food and Drug Administration restricted the

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use of silicone breast implants, and the Institute of Medicine issued a report¹ on the safety of silicone breast implants, citing local and perioperative complications as the principal safety issues, noting that risks mount over the lifetime of an implant. Based on the Institute of Medicine conclusion that there was no support for a novel syndrome associated with silicone breast implants, nor increased risk for cancer, connective tissue diseases, neurologic diseases, or other systemic complaints,¹ the Food and Drug Administration is considering reinstitution of silicone breast implants for augmentation mammoplasty.

The literature on existing silicone implants indicates as many as one third of symptomatic patients have ruptured gel implants at the time of explantation.² More than 465,000 women undergo chest surgery in the United States each year,³ with the potential for silicone from ruptured implants to be introduced into the pleural space at the time of surgery either through the surgery itself or chest tube placement. We present a case of fibrothorax due to introduction of silicone from ruptured implants at the time of atrial septal defect repair.

CASE REPORTS

A 61-year-old woman presented for evaluation of a hard right inframammary mass. She noted the onset of the mass approximately 1 year earlier, 1 year after cardiac surgery for closure of an atrial septal defect. The patient complained of discomfort around the mass and a vague sense of discomfort in the right hemithorax with respiration. She had no systemic complaints. Her history was significant for deep venous thrombosis, nephrolithiasis, and a distant history of pneumonia. Significant surgical history included bilateral mastectomies for severe fibrocystic disease with submuscular silicone gel implants for breast reconstruction, and hysterectomy. She never smoked and had no known exposure to asbestos. Physical examination demonstrated a hard, 3-cm mobile mass over the tenth rib in the midclavicular line, but was otherwise unremarkable. A chest radiograph revealed right pleural based masses, confirmed on chest CT. These were suspicious for mesothelioma or metastatic disease, and the patient underwent thorascopic biopsy.

Routine glass-mounted histologic sections stained with hematoxylin-eosin were examined and photographed by light microscopy. They were reprocessed for scanning electron microscopy (EM) and energy-dispersive radiograph elemental analysis (EDXEA) by the method of Pickett et al.⁴ The tissue sections were transferred to graphite specimen mounts, coated with carbon, and examined in an electron microscope (100-C TEM-SCAN; JEOL USA; Peabody, MA). Areas of interest studied by light microscopy were located and analyzed using a Tracor TN 5500 microprobe (Tracor-Northern; Madison, WI).

RESULTS

By light microscopy, hematoxylin-eosin-stained sections of the formalin-fixed, paraffin-embedded tissue showed lung parenchyma and overlying pleura, fibrous adhesions, and fibroadipose tissue. Within the pleura and fibroadipose tissue were numerous vacuoles of various sizes, occasionally surrounded by foreign-body giant cells. Translucent, refractile material was observed in many of the vacuoles (Fig 1).

By EM, the refractile material within the vacuoles appeared fluid and noncrystalline (Fig 2, *top*). EDXEA of

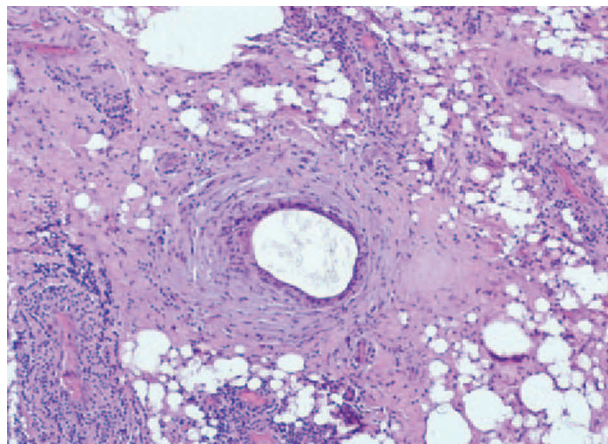


FIGURE 1. Light microscopy showing translucent, refractile material in many of the vacuoles (hematoxylin-eosin, original \times).

the material in the vacuoles showed the mineral content to be exclusively silicon (Fig 3). Dot mapping indicated that silicon distribution corresponded to the material seen in the light vacuoles by light microscopy (Fig 2, *bottom*).

Subsequently, the patient had both gel implants removed with free rupture of silicone noted at the time of surgery. The submammary mass was removed and found to be due to extruded silicone. The pleural masses have remained stable, although the patient notes pleuritic discomfort with painful respiration. Pulmonary function remains normal as documented by serial pulmonary function testing.

DISCUSSION

The frequency of rupture of gel implants is unknown. Brown et al⁵ found that 77% of women with silicone breast implants, without regard to complaints or symptoms, had at least one breast implant rupture; median implant age at the time of rupture was 10.8 years. Extruded silicone causes localized and distant areas of inflammation in the breast and surrounding tissues, including axillary lymph nodes, leading to the formation of pseudotumors.⁶⁻⁷ Diagnosis of ruptured implants is difficult and is performed with physical examination, mammography, ultrasound, CT, and MRI. None of these techniques can detect all ruptures; CT and MRI detect approximately 80% and 90%, respectively.^{8,9} Proof that a lesion is due to silicone requires further testing. We used a combination of EM and EDXEA to prove that silicon was contained in the pleural nodules. EDXEA has been used infrequently in medicine to determine elemental content of foreign material within tissue.

To our knowledge, this is the first reported case of fibrothorax due to the introduction of silicone from ruptured breast implants at the time of cardiac surgery. Though this patient's pulmonary function remains intact, fibrothorax can cause disabling dyspnea, and a severe restrictive defect or trapped lung, and may possibly require pleural decortication. We suspect the introduction of silicone into the pleural space in this case was associated with chest tube placement related to cardiac surgery. One case of pleural effusion was reported

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