Localized Fibrous Mesothelioma in the Postpartum Period

Thomas C. Rosenthal, MD
Buffalo, New York

Mesotheliomas have received much attention in the last decade as a result of their association with asbestos. A rare subgroup of pleural tumors was described in 1952 and named localized fibrous mesotheliomas.[1] These tumors were noted to originate from tissue beneath the superficial lining of the pleura (submesothelial connective tissue) as opposed to the more common type of mesothelioma that arises from the mesothelial lining itself and spreads diffusely. The localized fibrous mesotheliomas rarely metastasize (one case in 20) and are amenable to surgical resection and cure with only occasional recurrence (one case in six).[2] These tumors have been reported to be found in patients aged from 5 to 87 years (mean 53.7 years) and are unrelated to asbestos exposure.[3] The case reported here is the first known to have a critical impact on the postpartum course of an otherwise normal pregnancy.

CASE REPORT

D.N. was a 27-year-old woman who presented to the office at nine weeks' gestation for routine pregnancy care. Her history revealed that she was working as a gas station attendant. She had no history of factory work or known exposure to dangerous materials including asbestos. She had smoked less than one-half pack of cigarettes a day but had stopped with the pregnancy. She was taking no medications but admitted to occasional sampling of several street drugs before the pregnancy. She denied dependency or recent use. She had a peptic ulcer and appendectomy in the remote past. At the age of 15 years D.N. underwent a Harrington rod insertion for scoliosis, at which time she had received blood transfusions. The Harrington rod broke after surgery but remained in place. Her only previous obstetric history was an elective abortion at eight weeks' gestation six months before her first office visit. Her physical examination was consistent with pregnancy at nine weeks and normal except for scoliosis. There was no clubbing of her fingers or abnormal pulmonary findings.

Her pregnancy was unremarkable except for an episode of monilial vaginitis treated with miconazole vaginal cream when symptoms became severe at 19 weeks' gestation. At 28 weeks' gestation D.N. was treated with amoxicillin for a urinary tract infection. She had a 34-lb weight gain, and she demonstrated no signs of preeclampsia.

At 41 weeks' gestation D.N. presented in labor after spontaneous rupture of membranes at home. After 13 hours of labor (15 hours since rupture of membranes), she delivered a robust, 8-lb 7-oz, baby girl with a one-minute Apgar of 9, and a five-minute Apgar of 9.

Seven hours after delivery D.N. developed severe right-sided pleuritic chest pain. Arterial blood sampling revealed a pH of 7.49, oxygen (PaO_2) of 10.3 kPa (77 mmHg) and carbon dioxide ($PaCO_2$) of 10.0 kPa (30 mmHg). Continuous drip intraveneous heparin was begun for the presumptive diagnosis of pulmonary embolism. Platelet counts and a prothrombin and partial thromboplastin time were normal before the start of heparin.

The next day a chest x-ray film and a ventilation-perfusion lung scan were consistent with several right-sided pulmonary infarcts, but the studies were difficult to interpret because of chest deformity from scoliosis. A possible right-sided pleural effusion was noted. The patient's condition stabilized, and her partial thromboplastin time was maintained consistently at 1.25 to 1.5 times control values. Three days postpartum she suddenly developed a recurrence of right-sided chest pain. Hypovolemic shock ensued, requiring intubation, fluid replacement, pressor agents, and transfusions with six units of packed red blood cells and two units of fresh frozen plasma. Heparin therapy was discontinued immediately and protamine was administered. Bloody exudate was confirmed in the right pleural space by thoracentesis. On the fourth postpartum day she underwent a thoracotomy. A large vascular mass found in the right posterior pleura could not be resected

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From the Department of Family Medicine, School of Medicine, State University of New York at Buffalo, Buffalo, New York. Requests for reprints should be addressed to Dr. Thomas C. Rosenthal, Department of Family Medicine, School of Medicine, SUNY at Buffalo, 1001 Humboldt Parkway, Buffalo, NY 14208.

because of diffuse adhesions to the visceral and parietal pleura. She continued to experience bleeding problems, developed respiratory distress syndrome, and died on the eighth postpartum day.

At autopsy a localized fibrous mesothelioma originating from the right posterior visceral pleura was confirmed. She had multiple pulmonary emboli bilaterally and thrombophlebitis of the pelvic veins. Cause of death was determined to be respiratory failure resulting from massive pulmonary infarction and adult respiratory distress syndrome. The pleural tumor was somewhat lobulated with areas of hemorrhage and necrosis. Histologically, it demonstrated mostly acellular areas with whorls of collagen and spindle-like cells. There were some areas of cystic change. Areas that were more densely cellular showed only occasional mitotic figures. There was no evidence of metastasis.

DISCUSSION

Mesothelial cells line the visceral and parietal pleura, forming a complete layer of flattened cells that are engaged in active transport of fluid and small particles. They are covered with a glycocalyx that has a strong affinity for mucopolysaccarides, making the surfaces slippery. Mesothelial cells are highly susceptible to damage from air, water, asbestos, silica, and other agents. When damaged, they swell and lift off. Rather than healing by growing in from the edges, as does epithelium, the surfaces heal by regeneration of new cells as a sheet filling in the gap. Persistent or severe damage results in fibrous adhesions, or the repair process can lead to uncontrolled mitotic activity resulting in tumor growth.[3] Ethylene oxide, nitrosamines, chronic inflammation, beryllium, asbestos crystals, and radiation have all been associated with mesothelioma. There have been numerous reports of mesotheliomas resulting from radiation implants and cancer therapy, and one case of a localized mesothelioma occurred in a region that was repeatedly irradiated from diagnostic x-ray studies for a localized patient complaint.[4]

A search of the literature reveals less than 200 reported cases of localized fibrous mesothelioma. The most common symptoms are mild dyspnea and vague chest discomfort. One third may complain of rheumatoid-like joint symptoms. Hypoglycemia, chills, and fever are common presenting complaints. Clubbing of the fingernails may be present. Loss of weight is rare, and there is usually a paucity of localizing physical findings. Chest x-ray findings are not specific, but a pleural effusion is present in one sixth of the cases. The tumors occur in the right side of the chest twice as often as in the left side of the chest, and in the visceral pleura three times as often as in the parietal pleura. Many patients are asymptomatic but present to surgery because of abnormal findings on chest roentgenograms.[1,2,5–7]

Surgery is generally not considered difficult, as many tumors are attached by a vascular pedicle. A partial pneumonectomy is required in only one fourth of the cases, though some of the larger tumors have several attachments by adhesions. The tumors may recur in one sixth of the cases, and when there is recurrence, survival is usually limited to two years from the recurrence discovery. Death is due to respiratory failure or bleeding complications in the more vascular tumors. One fifth of the reported cases were diagnosed at autopsy.[1]

Gross features of these tumors are encapsulation with lobulation and an appearance suggestive of placental tissue. Histologically, they are highly variant tumors. The major cell type is the spindle cell arranged in areas that are cellular and areas that are sparsely cellular with dense whorls of collagen. There are usually areas with a cystic glandular appearance, and one third will have areas that are densely vascular, resembling hemangiopericytomas. The tumors may be covered with normal-appearing mesothelium.[3] All histologic arrangements were found in this patient. Except in the obviously malignant tumors, there are fewer than ten mitotic lesions per high-powered field.[1,2] Mesothelioma is a misnomer, as these tumors are distinct from the more common malignant mesothelioma.

SUMMARY

The case presented here is typical of the localized fibrous mesotheliomas reviewed in the literature. It had a disastrous impact on an otherwise unremarkable pregnancy. There is no discussion in the literature of humeral factors related to localized fibrous mesotheliomas, but the fever, chills, and rheumatoid complaints in some patients may suggest such factors. The presence of the tumor in the already hypercoagulable state of pregnancy may have predisposed this patient to pulmonary emboli. Anticoagulation led to hemorrhage from the thin-walled vascular areas of the tumor. This acute bleeding produced the hypovolemic shock on the third postpartum day, with a cascade of events leading to adult respiratory distress syndrome and the patient's ultimate death. This case underscores the importance of broad differential diagnosis as a continuing challenge in the practice of medicine.

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