Adenoid Cystic Carcinoma of Breast:

A Case with Recurrence and Regional Metastasis

WILLIAM B. WILSON, M.D., JAMES P. SPELL, M.D.

From the Departments of Pathology and Surgery, Mississippi Baptist Hospital, Jackson, Mississippi

ADENOID cystic carcinoma of the breast is very rare; its natural history has not been fully elucidated due to the paucity of reported cases. The literature contains 27 case reports, although only a few have been reported in detail. The present case is apparently the second with recurrence and metastases.

The tumor has been called cylindroma (Billroth [2]), adenocystic basal cell cancer of the breast (Geschickter [6]), adenocystic carcinoma of the breast (Foote and Stewart [4]) and adenoid cystic carcinoma of the breast (Ackerman [1]). In the breast, it is histologically identical with adenocystic carcinoma (cylindroma) of salivary glands and the respiratory tract where it is of low-grade malignancy, widely infiltrative, with a tendency to recur after operation and irradiation. From the literature [1-10] it is of low grade malignancy in the breast also, but is capable of producing metastases and probably death.

Case Report

In 1952, a 54-year-old married woman, who had two children, was seen because of a tumor in the right breast which had been present for "several" months. During this time she received hormonal therapy, but the tumor did not decrease in size. There was a mass about 5 cm. in diameter 5 cm. lateral to the nipple of the right breast without retraction, nipple discharge, or palpable axillary or supraclavicular nodes. Biopsy showed adenocystic basal cell carcinoma, the diagnosis by Dr. Kenneth Heard. Two days later, simple mastectomy was done. (The decision to do a simple mastectomy was influenced by the fact that none of Geschickter's four cases metastasized.)

Submitted for publication March 20, 1967.

On pathological examination the tumor was 2.8 cm. in diameter, and consisted of opaque gray tissue. On cross section, small cystic spaces were visible, from the walls of which papillary buds protruded. Microscopically, the tumor consisted of small, darkly staining cells, with slightly vesicular nuclei and scant cytoplasm. There was little pleomorphism, and nucleoli and mitotic figures were absent. The cells were disposed as anastomosing cords, narrowly separated by acellular zones of pink, hyaline material. When seen on cross-section, the cells had an acinar-like arrangement, with pink, acellular, hyaline material in the center of acini occasionally. Around the main tumor mass, there were small, well-circumscribed islands of similar tumor, infiltrating the fat of the breast tissue in characteristic manner (Fig. 1).

The patient did well until 1957, when a nodule appeared in the lateral portion of the healed incision. This was $2 \times 2 \times 3$ cm., and was stony hard and very tender. No axillary lymph nodes were palpable. The tumor was excised, and after frozen section diagnosis of carcinoma was made, the pectoral muscles and axillary contents were removed *en bloc*. Histologically, the tumor was identical with the previously excised carcinoma, although it was more cellular, and infiltration of perineural lymphatics was noted (Fig. 2). No lymphoid tissue was demonstrable around the tumor, and it was considered to be a local recurrence and not lymph node metastasis. The axillary lymph nodes were negative for tumor.

The patient continued well until 1967, when she was re-admitted for excision of a mass on the right lateral chest wall which had been present for 1 month. This appeared to be 3×5 cm. in size, nodular, slightly tender, and fixed to the chest wall. There was no evidence of tumor elsewhere, and chest, skull, and spine x-ray films were negative. At operation the tumor was attached to the periosteum of the 4th and 5th ribs, and extended upward into the axilla. The mass on the chest wall was excised, but it was not possible to remove all tumor from the axillary structures. On pathological examination, the tumor was $3.5 \times 7 \times 10$ cm. It was firm, discrete but not encapsu-

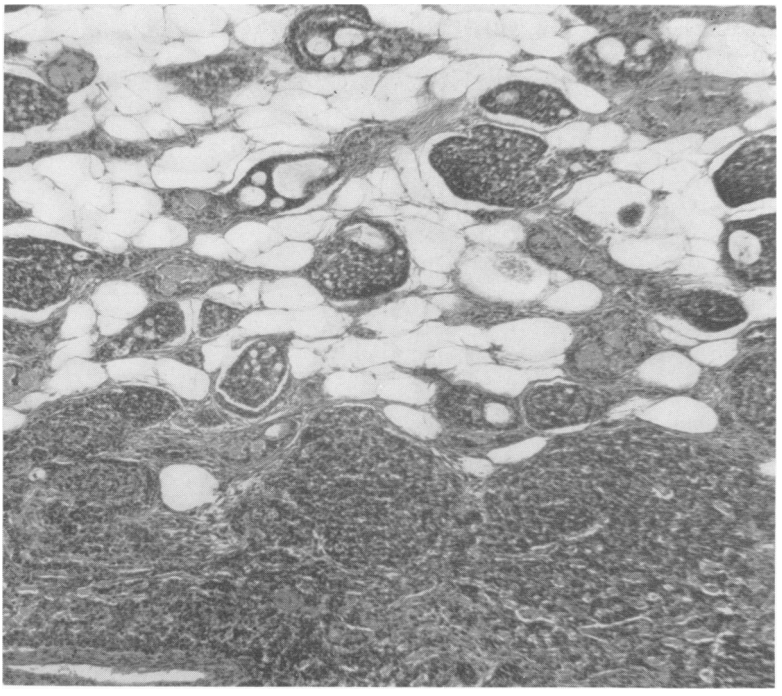

FIG. 1. Primary tumor. Circumscribed islands of cells infiltrate the fat around the main tumor mass, in a characteristic manner. Within the islands, spaces filled with faintly visible, hyaline, acidophilic material are seen. (40×)

lated, multilobular, and homogeneously yellow-pink. Grossly, it did not appear to invade surrounding tissue, and was well below the overlying ellipse of excised skin, which was not invaded. On sectioning, the tumor was homogeneously yellow-pink, and fleshy, with a central focus of cystic breakdown 1 cm. in diameter. Histologically, the tumor was typical of adenoid cystic carcinoma

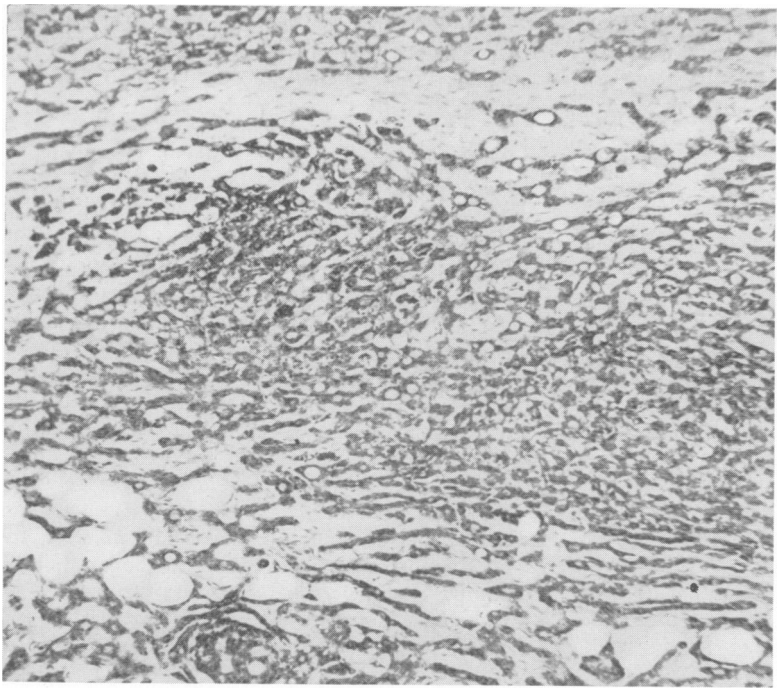

FIG. 2. First recurrence. A trabecular pattern is evident, again with characteristic hyaline material between the trabeculae and within the acinus-like clusters of cells. (40×)

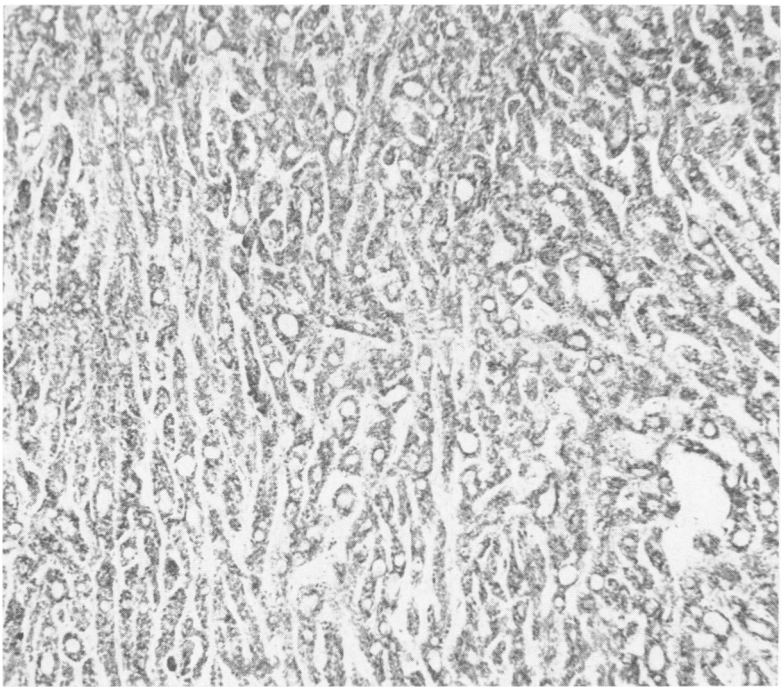

FIG. 3. Second recurrence. The same pattern is present. (100×)

and resembled previously excised tissue, although it was more cellular. In some areas the trabeculated appearance was lost and mitotic figures were abundant, although no pleomorphism was present (Fig. 3, 4). Along the edge of the tumor, a few scattered cells could be seen invading surrounding connective tissue, and a small vein in the fat near the edge of the tumor was filled with tumor cells.

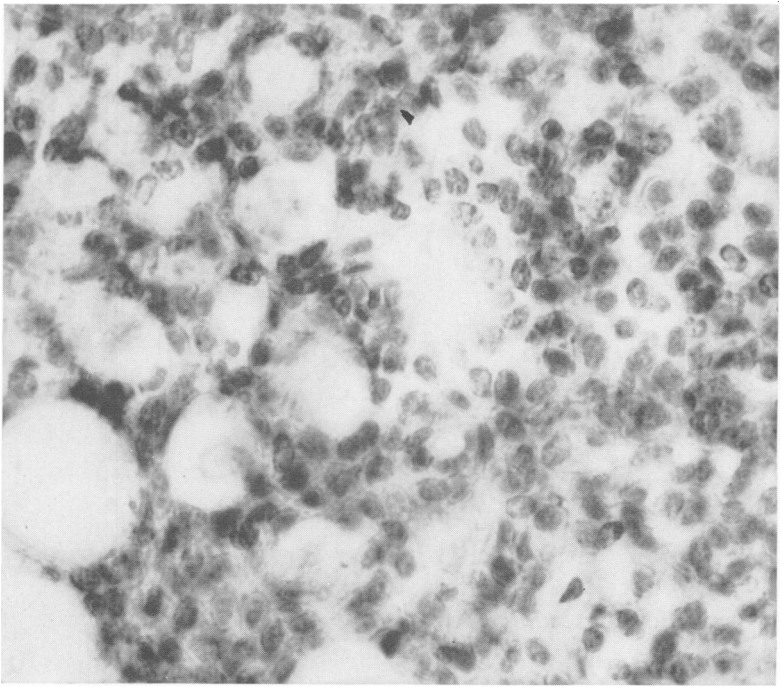

FIG. 4. Second recurrence. Well-differentiated cells with scant cytoplasm, in acinus-like arrangement, with hyaline material in the centers of the acinus-like spaces. (400×)

Perineural lymphatic invasion was also noted. The patient made an uneventful recovery from the operation.

Discussion

Galloway *et al.*[5] recently reviewed experience with this tumor in the breast over a 40-year period at the Mayo Clinic. They found nine examples. The tumors had been known to be present for up to 15 years at the time of excision, and were treated by operations varying from local excision to simple mastectomy to radical mastectomy. In no case were recurrence or metastases found in follow up from 1 to 17 years. In no case were axillary lymph node metastases found on pathologic examination, when lymph nodes were removed.

Although the Mayo Clinic experience indicates a favorable prognosis, the authors concurred with Ackerman[1] in believing that radical mastectomy is the most prudent therapy, pending further elucidation of the natural history of the lesion. The same opinion has been expressed in recent case reports.[3, 7] Only one case has been reported in which the tumor appears to have metastasized. This was the case of Nayer,[10] whose patient, at age 39, underwent radical mastectomy for a tumor which had been present for a year. The diagnosis of adenoid cystic carcinoma was made by Drs. Paul Klemperer and James Ewing. Axillary lymph nodes were negative for tumor. Eight years later, x-ray showed pulmonary nodules, but the patient lived 4 more years. At death she was cachectic, with pulmonary infiltration and pleural effusion. Cytologic examination of pleural fluid showed malignant cells in clusters, morphologically suggestive of adenoid cystic carcinoma of breast. These are illustrated in Nayer's report, but no biopsy or autopsy was done to prove the nature or origin of the metastatic carcinoma.

The present case, therefore, is the second reported of adenoid cystic carcinoma of the breast in which recurrence and regional metastases occurred. It is the first reported case with histologic confirmation of the recurrence by biopsy. This case also followed a prolonged course; 16 years. However, the experience indicates that adenoid cystic carcinoma is as capable of malignant behavior when primary in the breast as it is when it occurs elsewhere. This patient was treated initially by simple mastectomy, followed 7 years later by excision of a locally recurrent nodule and radical axillary lymph node dissection, followed by another recurrence 15½ years after initial mastectomy.

Summary

Adenoid cystic carcinoma is a rare form of cancer in the breast of low grade malignancy. However, the case reported indicates that the tumor is capable of widespread local recurrence, and a previously reported case indicates that the tumor was responsible for pulmonary metastasis and death. Experience with this case supports the current consensus that radical mastectomy is the treatment of choice.

References

1. Ackerman, L. V.: Surgical Pathology (3rd Ed.). St. Louis, C. V. Mosby Co., 1964.
2. Billroth, T.: Die Cylindergeschwalst. Untersuchungen ueber die Entwicklung der Blutgefasse. Berlin, G. Reimer, 1856.
3. Eufemio, G. and Villaflor, V. V.: Adenoid Cystic Carcinoma (Cylindroma) of Breast. Acta Med. Philipp., 1:212, 1965.
4. Foote, R. W., Jr. and Stewart, F. W.: A Histologic Classification of Carcinoma of the Breast. Surgery, 19:74, 1946.
5. Galloway, J. R., Woolner, L. B. and Claggett, O. T.: Adenoid Cystic Carcinoma of the Breast. Surg. Gynec. Obstet., 122:1289, 1966.
6. Geschickter, C. F.: Diseases of the Breast (2nd Ed.), Philadelphia, J. P. Lippincott Co., 1945.
7. Groshong, L. E.: Adenocystic Carcinoma of Breast. Arch. Surg., 92:424, 1966.
8. Leonardelli, G. B. and Pizzetti, F.: I cilindromi. Arch. Ital. Otol., 64:318, 1953.
9. Lewison, E. F.: Breast Cancer and Its Diagnosis and Treatment. Baltimore, Williams & Wilkins Co., 1955.
10. Nayer, H. R.: Cylindroma of the Breast with Pulmonary Metastases. Dis. Chest, 31:324, 1957.