Invasive Ductal Carcinoma in a Mammary Hamartoma: Case Report and Review of the Literature

Nami Choi, MD¹
Eun Sook Ko, MD²

Mammary hamartomas are typically a benign condition and rarely develop into malignant lesions. Only 14 cases of carcinomas associated with a hamartoma have been documented in the literature. In this case report, we describe a case of invasive ductal carcinoma within a hamartoma in a 72-year-old woman. Mammography, ultrasonography, and magnetic resonance imaging showed the features of a typical hamartoma with a suspicious mass arising in it. This case illustrates the importance of identification of unusual findings in a typical mammary hamartoma on radiologic examinations.

CASE REPORT

A 72-year-old woman presented for further examination of a right breast lump. According to the patient, the lump had been present for at least 10 years. She complained of discomfort of recent onset in the right breast. On physical examination, a soft, mobile, 10 cm mass, which occupied nearly the entire right breast, was palpated. On mammography, a large circumscribed mass surrounded by a water-density capsule was shown in the right breast (Fig. 1A, B). The mass was a mixture of isodense and fat densities and had dystrophic calcifications in a branching pattern. The mammograms were initially interpreted as a benign hamartomatous lesion (Fig. 1A, B). On ultrasonography (US), the mass was very heterogeneous and completely encompassed in a thin echogenic pseudocapsule of compressed breast tissue, which was compatible with hamartoma. However, a careful US examination revealed an irregular hypoechoic mass of 1.4 cm with a non-parallel orientation within the hamartoma (Fig. 1C). A retrospective review of the mammograms revealed focal asymmetry, which correlated with the suspicious mass on US (Fig. 1A, B). The lesion was early enhanced and a washout on enhanced MRI scans (Fig. 1D, E), thus it was
thought to be a malignancy. Surgical excision for entire mass was done and the diagnosis from the frozen specimen of the suspicious area was IDC.

The patient therefore underwent a modified radical mastectomy. The microscopic examination showed an IDC arising in a hamartoma (Fig. 1F–H). There was no IDC which was outside the hamartoma. There was no intraductal component and there was no axillary lymph node metastasis. The remaining palpable mass was hamartoma with dystrophic calcifications. The patient subsequently underwent adjuvant chemotherapy. At 24 months post surgery, there was no evidence of local recurrence of the IDC or any distant metastasis.

**DISCUSSION**

Mammary hamartomas, a term applied to breast tumors in 1971 by Arrigoni et al. (13), have also been referred to as lipofibroadenomas, adenolipofibromas, and fibroadenolipomas (1). Hamartomas may present as tender or non-

| Table 1. Summarized Clinicopathologic Features of Reported Hamartoma-Associated Malignancy |
|-----------------------------------------|------------------------------------------|-----------------|-----------------|-----------------|-----------------|
| Age | Clinical Findings | Mammographic Findings | US Findings | Histology (Hamartoma / Carcinoma) | Size (cm) |
|-----|------------------|----------------------|-----------|---------------------------------|----------|
| Mendiola et al., 1982 (3) | 54 | Painful lump | Typical hamartoma with several clusters of microcalcifications | No information | LCIS | 6 cm/no information |
| Coyne et al., 1992 (5) | 59 | MMG detected, soft palpable mass | Typical hamartoma with no suspicious feature | No information | ALH, LCIS and microILC | IDC extending to skin | 7 cm/3 cm |
| Anani et al., 1996 (11) | 78 | Soft, palpable mass with skin ulcer | Typical hamartoma with irregular opacity containing microcalcifications | No information | IDC | 5 cm/2 cm |
| Anani et al., 1996 (11) | 53 | Soft, palpable mass | Typical hamartoma with spiculated mass | Hypoechoic absorbing lesion suggesting carcinoma | IDC | 5 cm/2 cm |
| Mester et al., 2000 (2) | 59 | Lump | Typical hamartoma with pleomorphic calcifications | No information | DCIS, LCIS and IDC | 7 cm/3.5 and 3.0 cm |
| Kuroda et al., 2002 (12) | 53 | Lump | Typical hamartoma with no suspicious findings | Typical hamartoma | MicroILC | 6 cm/0.3 cm |
| Tse et al., 2002 (10) | 32 | Lump | Not performed | Well-defined lobulated mass, suggesting benign carcinoma | DCIS | 1.5 cm/0.7 cm |
| Tse et al., 2002 (10) | 68 | Lump | Not performed | No information | DCIS, mucinous ca | 2.5 cm/0.8 cm DCIS, 0.1 cm mucinous ca |
| Lee et al., 2003 (7) | 65 | Axillary node, no breast mass palpable | Typical hamartoma with spiculated density containing microcalcifications | Well-defined hypoechoic nodule within hamartoma | DCIS and IDC | 6 cm/1 cm IDC |
| Lee et al., 2003 (7) | 68 | MMG detected, soft palpable mass | Typical hamartoma with spiculated mass | No information | IDC | No information/2 cm |
| Baron et al., 2003 (4) | 54 | Known hamartoma, soft mobile mass | Typical hamartoma with fine pleomorphic microcalcifications | No information | ILC | 5 cm/2 cm |
| Ruiz-Tovar et al., 2006 (9) | 35 | Lump | Typical hamartoma with grouped microcalcifications | No information | DCIS and IDC | No information/1.2 cm IDC |
| Kai et al., 2009 (6) | 70 | Known hamartoma, firm, ill-defined, mobile, spherical tumor | Typical hamartoma grouped amorphous calcifications and distortion | Irregular hypoechoic tumor, both within and external to hamartoma | DCIS and IDC | 3.2 cm/3.5 cm |
| Pervatikar et al., 2009 (8) | 25 | Gradual growing lump | Not performed | No information | IDC | 12 cm/1.5 cm |
| Choi et al. (this study) | 72 | Lump for at least 10 years with discomfort of recent onset | Typical hamartoma with focal asymmetry | Spiculated non parallel hypoechoic nodule within hamartoma | IDC | 9 cm/1.5 cm |

Note. — ALH = atypical lobular hyperplasia, DCIS = ductal carcinoma in situ, IDC = invasive ductal carcinoma, ILC = invasive lobular carcinoma, LCIS = lobular carcinoma in situ, MMG = mammography
tender palpable lumps, but are often discovered incidentally during a screening mammography (1, 14). The typical mammographic feature of hamartomas is a circumscribed fibrofatty mass (15). On US, most mammary hamartomas have circumscribed margins, an oval shape, and heterogeneous internal echogenicity (16, 17).

Malignancies associated with hamartomas are rare. The clinical, radiologic, and histologic findings of the previously described 14 cases of malignant hamartomas and the current case are summarized in Table 1. The mean patient age was 56.3 years (range, 25–78 years). The lesions are frequently recognized as palpable masses. The size of the hamartomas range from 1.5–12.0 cm in diameter and the size of the associated carcinomas range from 0.3–3.5 cm in diameter.

Of the described 15 cases, mammography was obtained in 12 cases, of which 10 showed the typical appearance of hamartomas with suspicious features, such as clusters of microcalcifications, pleomorphic micocalcifications, and spiculated masses. The remaining two cases had the typical

![Fig. 1. Invasive ductal carcinoma and mammary hamartoma in 72-year-old woman. A, B. Mediolateral oblique and craniocaudal views of right breast show fat-containing mass including dystrophic calcifications, suggesting hamartoma. There is focal asymmetry (arrows) at 12 o’clock within hamartoma. C. US of right breast at 12 o’clock shows spiculated, nonparallel, hypoechoic mass, which corresponds with focal asymmetry on mammogram (A, B). D, E. Standard subtraction image (D) and reverse subtraction image (E) of dynamic MRI show suspicious mass (arrows) within hamartoma, which was early enhanced (D) and washout (E). F. Gross specimen shows 9-cm smooth circumscribed fatty mass. Suspicious mass was excised from fatty mass (arrow).](image-url)
appearance of a hamartoma with no suspicious features, thus co-existing malignancies were unexpected findings at the time of tumorectomy. US findings were available in only six cases, of which four had suspicious masses with irregular margins, hypoechochogenicity, or a non parallel orientation within the hamartomas; and two cases were diagnosed pre-operatively as carcinomas by US-guided fine needle aspiration or core needle biopsies and underwent one-step curative surgery (6, 7). As stated above, the majority of cases had suspicious findings within the hamartoma on mammography or US. Radiologists therefore need to pay careful attention in order to detect subtle suspicious findings, even though mammography or US may show typical hamartomas.

Among the 15 cases described here, 12 had carcinomas that were confined to the hamartomas and the remaining three cases had carcinomas that involved both the hamartomas and adjacent normal breast tissue. If carcinomas involve both the hamartoma and normal breast tissue, it is difficult to determine whether the carcinoma arises within the hamartoma or an isolated carcinoma which initiates growth nearby later extends into the hamartoma. However, in the majority of cases described here, including the current case, the carcinomas were located within the hamartomas, (2–5, 7, 8, 10–12) thus we believe that the malignancies arose within the hamartomas.

In conclusion, breast hamartomas have generally been classified as rare, benign tumors, and carcinomas occur only rarely. However, hamartomas may be diagnosed with greater frequency due to widespread screening mammography. Radiologists should recognize that malignancy may co-exist or develop in hamartomas and be alert to the presence of suspicious features within a hamartoma.

References
1. Stavros AT. Breast ultrasound. Philadelphia: LWW, 2004:560-569
2. Mester J, Simmons RM, Vazquez MF, Rosenblatt R. In situ and infiltrating ductal carcinoma arising in a breast hamartoma. AJR Am J Roentgenol 2000;175:64-66
3. Mendiola H, Henrik-Nielsen R, Dyreborg U, Blickert-Toft M, Al-Hariri JA. Lobular carcinoma in situ occurring in adenolipoma of the breast. Report of a case. Acta Radiol Diagn (Stockh) 1982;23:503-505
4. Baron M, Ladonne JM, Gravier A, Picquenot JM, Berry M. Invasive lobular carcinoma in a breast hamartoma. Breast J 2003;9:246-248
5. Coyne J, Hobbs FM, Boggis C, Harland R. Lobular carcinoma in a mammary hamartoma. J Clin Pathol 1992;45:936-937
6. Kai M, Tada K, Tamura M, Gomi N, Horii R, Akiyama F, et al. Breast cancer associated with mammary hamartoma. Breast Cancer 2009 [Epub ahead of print]
7. Lee EH, Wylie EJ, Bourke AG, Bastiaan De Boer W. Invasive ductal carcinoma arising in a breast hamartoma: two case reports and a review of the literature. Clin Radiol 2003;58:80-83
8. Pervatikar SK, Rao R, Dinesh US, Parameswararai A S. Large mammary hamartoma with focal invasive ductal carcinoma. Indian J Pathol Microbiol 2009;52:249-251
9. Ruiz-Tovar J, Requero-Callejas ME, Aliáz AB, Ramiro C, Rojo R, Collado MV, et al. Infiltrating ductal carcinoma and ductal carcinoma in situ associated with mammary hamartoma. Breast J 2006;12:368-370
10. Tse GM, Law BK, Pang LM, Cheung HS. Ductal carcinoma in situ arising in mammary hamartoma. J Clin Pathol 2002;55:541-542
11. Anani PA, Hessler C. Breast hamartoma with invasive ductal carcinoma. Report of two cases and review of the literature. Pathol Res Pract 1996;192:1187-1194
12. Kuroda N, Sugimoto T, Numoto S, Erzan H. Microinvasive...
lobular carcinoma associated with intraductal spread arising in a mammary hamartoma. J Clin Pathol 2002;55:76-77

13. Arrigoni MG, Dockerty MB, Judd ES. The identification and treatment of mammary hamartoma. Surg Gynecol Obstet 1971;133:577-582

14. Daya D, Trus T, D’Souza TJ, Minuk T, Yemen B. Hamartoma of the breast, an underrecognized breast lesion. A clinicopathologic and radiographic study of 25 cases. Am J Clin Pathol 1995;103:685-689

15. Helvie MA, Adler DD, Rebner M, Oberman HA. Breast hamartomas: variable mammographic appearance. Radiology 1989;170:417-421

16. Chao TC, Chao HH, Chen MF. Sonographic features of breast hamartomas. J Ultrasound Med 2007;26:447-452

17. Park SY, Oh KK, Kim EK, Son EJ, Chung WH. Sonographic findings of breast hamartoma: emphasis on compressibility. Yonsei Med J 2003;44:847-854