A Rare Case of Recurrent Myoid Hamartoma Mimicking Malignancy: Imaging Appearances

Myung-Su Ko, MD
Won Sang Jung, MD
Eun Suk Cha, MD
Hyun Joo Choi, MD

Myoid hamartoma is an uncommon type of breast hamartoma and its recurrence is very rare. We report the imaging appearance of an unusual case of recurrent myoid hamartoma of the breast mimicking malignancy in a 43-year-old woman. Although the mammographic and ultrasonographic findings have long been described in the literature, MR finding with a dynamic study has not, to the best of our knowledge, been reported previously.

Breast hamartomas are rare breast neoplasms, with the main characteristic being the various tissues they contain. Histologically, breast hamartomas have been classified as various types based on the predominant type of tissue, which include fibrous, fibrocystic, and fibroadenomatous tissue with adipose tissue. Myoid hamartoma which has smooth muscle cells is a rare type (1). As the recurrence of a myoid hamartoma is very rare, we report a patient with recurrent myoid hamartoma of the breast, describe the radiologic findings including MR, and review the relevant literature.

CASE REPORT

A 43-year-old woman had undergone surgery for a myoid hamartoma in the lower outer quadrant of the patient’s left breast. Twelve months later, she presented with a palpable lump under the surgical scar. A clinical breast examination revealed a 3.0-cm sized firm mass with slight mobility under the scar. A mammography showed two oval isodense masses with a partially obscured margin in the left subareolar area (Fig. 1A). Ultrasound images showed a 2.3-cm sized irregular isoechoic mass with a microlobulated margin situated in the surgical bed (Fig. 1B). Another 2.0-cm sized, oval hypoechoic mass with a circumscribed margin was found in the lower outer quadrant of the patient’s left breast (Fig. 1C). US examination results indicated that the mass in the surgical bed was considered to be suspicious. To further evaluate the suspicious mass, MRI was performed on a 1.5 Tesla system (General Electric Medical Systems, Milwaukee, WI) with a specially developed double breast coil. The MR revealed two, circumscribed, oval-shaped masses which appeared to be of isosignal intensity relative to skeletal muscle on the T1-weighted images (WI) and with high-signal intensity on the T2-WI. On contrast enhanced T1-WI and subtracted images, the masses showed strong enhancement. On dynamic MRI, the mass situated in the surgical bed showed early intense enhancement and progressive signal loss over time (Fig. 1D, E). This finding was considered to be suspicious for malignancy. The lesion found in the lower outer quadrant of the left breast showed progressive enhancement (Fig. 1F, G) and...
thought to be benign.

A 14-gauge core needle biopsy was performed on the mass under the scar using ultrasound guidance. A microscopic evaluation revealed that the lobules and ducts of the mammary gland and stroma had no malignant component and reported to be a fibroadenoma. The patient requested and then underwent a surgical excision in order to obtain a definitive diagnosis. The gross pathology evaluation of the specimen revealed a $2.5 \times 1.8 \times 2.0$-cm-long rubbery, white, solid mass situated in the surgical bed and a $2.3 \times 1.4 \times 2.4$-cm-long rubbery, white, solid mass in the lower outer quadrant of the left breast; both masses were round and had smooth margins. The microscopic evaluation of the mass situated in the surgical bed revealed lobules and ducts of the mammary gland, stroma with a high collagen concentration, and many

![Fig. 1. 43-year-old female patient.](image)

A. Medio-lateral oblique view of left breast shows two oval isodense masses (arrows and arrowheads) with partially obscured margin (arrows) in left subareolar area.
B, C. Ultrasound images show 2.3-cm sized irregular isoechoic mass (white arrowheads) with microlobulated margin located between surgical scars (arrows) and overlying skin scar (black star B), and another, 2.0-cm sized oval hypoechoic mass with circumscribed margin located in lower outer quadrant of left breast (C).
D. Contrast-enhanced sagittal subtracted MR image reveals rapidly enhancing mass.
bundles of smooth muscle fiber without atypia (Fig. 1H). An immunohistochemical investigation was positive for vimentin, smooth muscle actin, CD34 in the smooth muscle cells, but negative for S100 protein. The final pathology diagnosis was myoid hamartoma, whereas the other mass was a fibroadenoma.

DISCUSSION

Breast hamartomas consist of approximately 0.7–5% of all benign breast tumors (2). They are rare benign breast neoplasms consisting of a disorganized overgrowth of normal breast tissue. Breast hamartomas are usually classified subtypes based on the predominant tissue. Myoid hamartoma is a rare hamartoma subtype characterized by the presence of histologically normal, but irregular, randomly distributed, smooth muscle cells (3).

There are several theories regarding the origin of smooth muscle cells (3, 4), which include that muscle cells originate from stroma via leiomyomatous metaplasia, local vessel

Fig. 1. 43-year-old female patient.
E. Graph of MRI time-signal intensity curves from MR image shows rapid enhancement on first postcontrast time point and significant drop in signal on subsequent time points.
F. Contrast-enhanced sagittal subtracted MR image reveals progressive enhancement of mass.
G. Graph of MRI time-signal intensity curves from MR image shows gradual enhancement on first postcontrast time point and continuous increase in signal on subsequent time points.
H. Photomicrograph shows smooth muscle fibers without atypia (Hematoxylin & Eosin stain, ×100).
walls, or myoepithelium via a metaplastic process.

A mammography usually shows a well-circumscribed mass with a radiolucent area, suggestive of fat tissue, and sometimes a thin capsule (5). Ultrasound often shows well-circumscribed masses of heterogeneous density and ranging from hypoechoic to hyperechoic (6).

Although breast hamartoma has long been described in the literature, there have been only a few descriptions of their MR appearance (7). A previous report demonstrates the MR characteristics of a hamartoma that had a high signal on T1WI and in which the moderate signal seen on T2WI corresponded to the fatty component of the mass. However, in this case, the myoid hamartoma showed no high signal on T1WI and showed early intense enhancement and progressive signal loss over time (washout) on dynamic MRI, which suggested malignancy (7). Therefore, arriving at a differential diagnosis from malignancy was difficult in this case. The fibroadenoma showed progressive enhancement, which made it more likely to be benign (7).

A core biopsy may be diagnostic for a myoid hamartoma, although a subsequent excisional biopsy and immunohistochemical evaluation ensure the definite diagnosis of a breast hamartoma (8). Microscopically, smooth muscle cell, fibrous stroma, and adipose tissue are evident. The immunohistochemistry examination revealed that the findings were positive for the presence of smooth muscle actin, desmin, as well as vimentin, and negative for S-100 protein (3).

Local excision is the treatment of choice for a myoid hamartoma, although no adjuvant therapy is needed following surgery. Although there are no data regarding the tumor recurrence rate, reported cases of tumor recurrence are very rare with only one case having been reported by Linell et al. (9). Our patient experienced recurrence is 12 months.

In conclusion, we have presented a rare case of recurrent myoid hamartoma in which the MR features mimicked malignancy.

References
1. Sharkey FE, Allred DC, Valente PT. Breast. In: Damjanovi I, Linder J, eds. Anderson's pathology. St. Louis: Mosby-Yearbook, 1996:2363-2364
2. Charpin C, Mathoulin MP, Andrac L, Barberis J, Boulat J, Sarradour B, et al. Reappraisal of breast hamartomas. A morphological study of 41 cases. Pathol Res Pract 1994;190:362-371
3. Stafyla V, Kotsifopoulos N, Grigoriadis K, Bakoyiannis CN, Peros G, Sakorafas GH. Myoid hamartoma of the breast: a case report and review of the literature. Breast J 2007;13:85-87
4. Di Tommaso L, Pasquinelli G, Damiani S. Smooth muscle cell differentiation in mammary stromo-epithelial lesions with evidence of a dual origin: stromal myofibroblasts and myoepithelial cells. Histopathology 2003;42:448-456
5. Ruiz Tovar J, Regueiro Callejas ME, Alaez Chillarón AB, Ramiro Pérez C, Collado Guirao MV, Rojo Blanco R, et al. Mammary hamartoma. Clin Transl Oncol 2006;8:290-293
6. Georgian-Smith D, Kricun B, McKee G, Yeh E, Rafferty EA, D’Alessandro HA, et al. The mammary hamartoma: appreciation of additional imaging characteristics. J Ultrasound Med 2004;23:1267-1273
7. Wiener JI, Schilling KJ, Adami C, Obuchowski NA. Assessment of suspected breast cancer by MRI: a prospective clinical trial using a combined kinetic and morphologic analysis. AJR Am J Roentgenol 2005;184:878-886
8. Rosser RJ. Epithelioid cells in myoid hamartoma of the breast. Arch Pathol Lab Med 1997;121:354-355
9. Linell F, Ostberg G, Söderström J, Andersson I, Hildell J, Ljungqvist U. Breast hamartomas. An important entity in mammary pathology. Virchows Arch A Pathol Anat Histol 1979;383:253-264