

Report

Report of 6 Cases of Mammary Hamartoma**Kei AOYAMA, Takako KAMIO, Tetsuya OHCHI,
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Mammary hamartoma, which is composed of fat, mammary gland, and connective tissue, to form a tumor mass, is classified as a tumor-like lesion since it is a nonneoplastic lesion. The image findings depict a tumor mass with flat margins having a well-defined border in all cases, so that it is difficult to distinguish the tumor mass from fibroadenoma. A radiolucent zone is observed in mammographic findings, while breast ultrasonographic findings consist of an oval or segmented shape with a well-defined border, while the internal echogenicity reveals various findings, including a uniform, tachetic, linear or marbled structure. In a magnetic resonance imaging (MRI), the tumor mass is depicted as having a well-defined border and flat margins, T1-enhanced images of the interior reveals satisfactory distinction with mammary gland tissue with fat components demonstrating well-defined, high signals on T1-weighted imaging. The radiolucent zone of the tumor mass is peculiar in a mammography (MMG) and the stripe marble pattern (dendriform) is peculiar in ultrasonography (US) and an MRI. The description rate of MMG was 82%, US was 33%, and the MRI was 100%. Because fat tissue was described in the clear high signals on T1-weighted imaging in the MRI, the internal mixture pattern was well described with the contrast with low-signal gland tissue most sharply shown. As the detection rate is 100% even for the fat element in a little tumor mass, the sensitivity is very high.

This is reportedly very rare and benign, but sometimes can become enormous, and breast cancer could potentially occur within the hamartoma. It can be accurately diagnosed and treated if the characteristic images are properly revealed.

Key words: breast, mammary hamartoma

Introduction

Examples of benign mammary tumor masses frequently encountered during routine examination include fibroadenoma, mastopathy and phyllodes tumor. Although mammary hamartoma is extremely rare, it can be distinguished from other benign tumors relatively easily since it exhibits a characteristic image. Mammary hamartoma was first proposed by Ariigoni et al¹⁾ in 1971 as a localized mammary tumor mass resembling fibroadenoma. Hamartoma is defined as a focal deformity resembling a neoplasm in which, although the site where various tissue elements are present is normal, the mixture of cells and tissue is abnormal or the composition of a single element is abnormal. We encountered 6 cases

of mammary hamartoma and report on the results with a brief bibliography.

Materials and Methods

From 2002 to 2006, a study was conducted at our hospital on the histopathologic diagnosis of the respective characteristic image findings obtained from mammography (MMG), ultrasonography (US) and magnetic resonance imaging (MRI) in 6 patients diagnosed as having hamartoma.

Results

The motivation for examination was the awareness of a tumor mass in all 6 patients. The duration of symptoms was more than 5 years in 3 patients, and less than 1 year in 3 patients. All patients were recognized to a tumor mass and 4 cases were over-

Table Image findings and pathologic diagnosis in six cases

Age (y.o)	Size (cm)	MMG findings		US findings		MRI findings		Diagnostic approach	Pathological diagnosis	
		internal density	radiolucent zone	shape	internal structure	shape	internal structure			
a	66	8	low	+	oval	heterogeneous	well-circumscribed, dendriform	heterogeneous, marbled	excision	A
b	26	8	low and high	+	oval, nodular	heterogeneous	well-circumscribed, nodular	heterogeneous, marbled	excision	F
c	58	5	central high/ peripheral low	+	oval	heterogeneous, marbled	–	–	excision	A
d	40	5	high	–	segmented, tachetic	heterogeneous	–	–	biopsy	F
e	46	8	central high/ peripheral low	+	oval	heterogeneous, marbled	well-circumscribed, dendriform	heterogeneous, marbled	biopsy	A
f	38	8	high	+	oval	heterogeneous	well-circumscribed, dendriform	heterogeneous, marbled	biopsy	A

A: adenolipoma (lipomatous hamartoma), F: fibroadenolipoma (fibroadenomatous hamartoma).

looked as breast asymmetry. There is also the possibility of small tumor masses included at routine outpatient examinations. Some cases are diagnosed as fibroadenoma as a result of being only partially sampled in cases diagnosed with needle biopsy. Although some cases are undiscovered hamartoma, a comparative study was conducted on 6 cases which were diagnosed on the basis of image findings and histopathologic diagnosis (Table). Patient ages, ranged from 26 to 66 years with mean of 45.7 years. There were no characteristics observed in the age distribution of the hamartoma cases we encountered. Although the image findings revealed circumscribed margins in all cases, the internal structures varied depending on whether the tissue was lipomatous hamartoma or fibroadenomatous hamartoma.

MMG were performed as a routine diagnostic test for breast screening in all 6 patients. All cases demonstrated well defined margins. Four cases (b, c, e, and f) showed heterogeneous density slightly higher than the surrounding mammary gland tissue. One case (d) showed high density, and another (a) was low density. Characteristic MMG findings consisted of circumscribed tumors demarcated by a radiolucent zone, and fat density within the tumor mass according to the types and proportions of constituent components, as in cases a, b, c, e and f (Fig. 1).

US findings revealed spherical or oval masses in

shape, and the heterogeneous internal echogenicity in all 6 cases (Fig. 2). The internal echogenicity was non-uniform due to the effect of internal constituent components of the fatty tissue and gland tissue.

MRI were performed in 4 patients, revealed a well-defined border in all cases, and the internal heterogeneous intensity with either nodular or dendriform enhancement (Fig. 3).

Cases a, b and c were diagnosed by surgical excisional biopsy, because of patient's preference. Cases d, e and f were diagnosed by core needle biopsy under US guidance. The macroscopical findings of excised specimens revealed capsules, cases a and c had much fat tissue in the constituent components exhibited a solid yellow color, with a grayish-white color partially contained in the tachetic area. Case b showed a nodular, grayish-white color with some tachetic yellow areas (Fig. 4).

Discussion

Mammary hamartoma are uncommon, benign, and slow growing: they are reported as 0.7% of all benign mammary tumors in women². The hamartoma is different from mastopathy among the benign mammary tumors in that the lesion is well-circumscribed, and different from fibroadenoma in terms of tissue structure and the containing of fat tissue. However, hamartomas are difficult to distinguish from phyllodes tumors in bigger lesions, and from fibroadenoma in smaller lesions. The lesions tend to easily become familiar to surrounding mam-

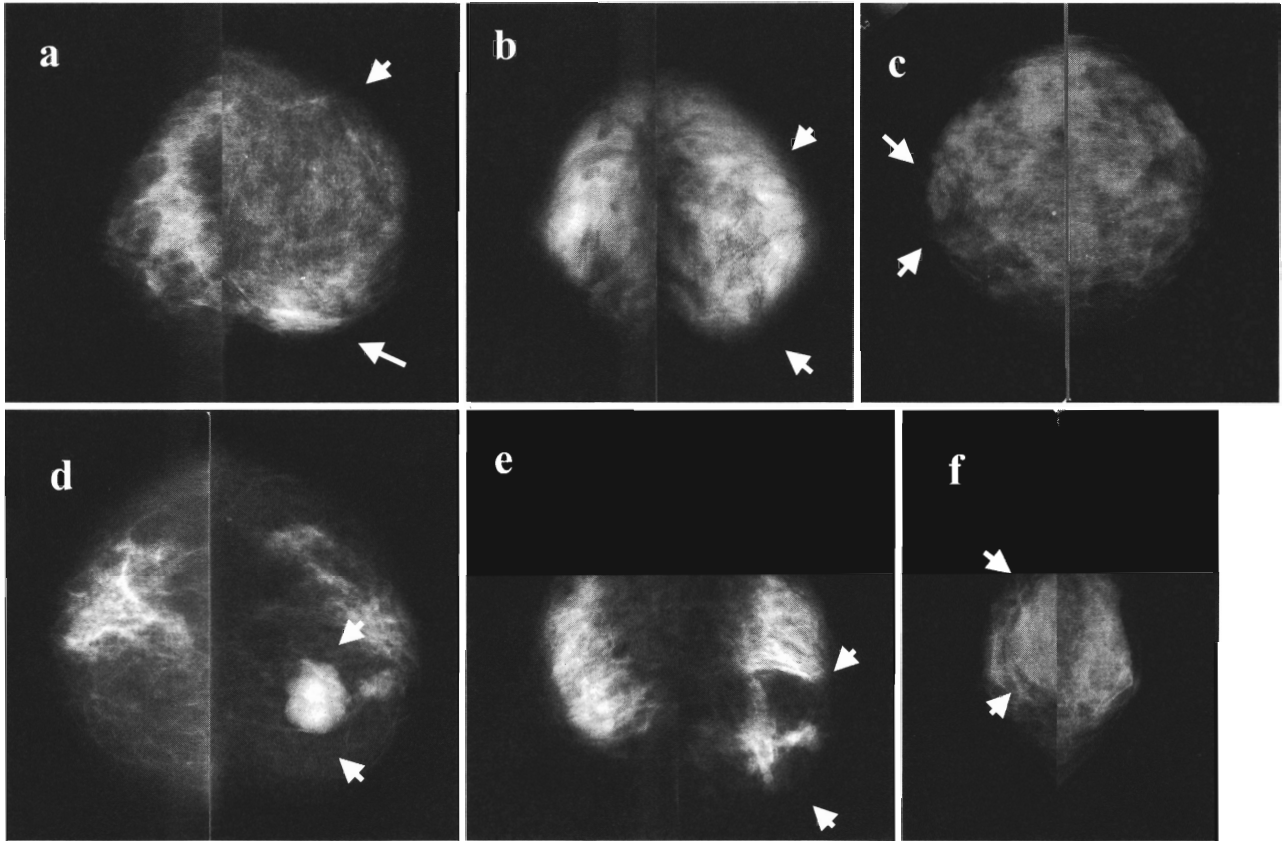


Fig. 1 Mammographic findings

a: case a. An oval tumor mass was observed in the left breast with a heterogeneous density and exhibiting a radiolucent zone.

b: case b. An oval tumor mass was observed in the left breast with a heterogeneous density and exhibiting a radiolucent zone.

c: case c. An oval tumor mass was observed in the right breast with a heterogeneous density and exhibiting a radiolucent zone.

d: case d. A segmented, high-density tumor mass was observed in the left breast having a well-defined border.

e: case e. An oval tumor mass was observed in the left breast having a heterogeneous interior density and exhibiting a radiolucent zone.

f: case f. An oval tumor mass was observed in the right breast having a heterogeneous interior density and exhibiting a radiolucent zone.

mary gland tissue, and some cases were not recognized to be tumor masses and were often overlooked as breast asymmetry, thus resulting in a long duration of symptoms. Hamartoma is a tumor mass with a well-defined border with surrounding tissue within the breast, and is composed of tissue that is either identical to tissue components of the breast or is missing a portion thereof. Moreover, the proportion of each tissue component differs considerably from normal tissue. Sakamoto et al classified subtypes of hamartoma as follows: (1) adenolipoma that is lipomatous overall while containing a small amount of mammary gland tissue therein (lipoma-

tous hamartoma); (2) fibroadenolipoma that is fibroadenomatous overall while containing fat tissue in a portion thereof (fibroadenomatous hamartoma); (3) chondrolipoma in which cartilaginous tissue is present along with glandular, fibrous and fat tissue (chondromatous hamartoma); and (4) myoid hamartoma in which smooth muscle tissue is similarly present.

Mammary hamartoma has no capsule and is composed of a mixture of gland tissue and fat tissue in varying proportions. The lesions tend to easily become familiar to surrounding mammary gland tissue and are not recognized as a tumor mass, thus

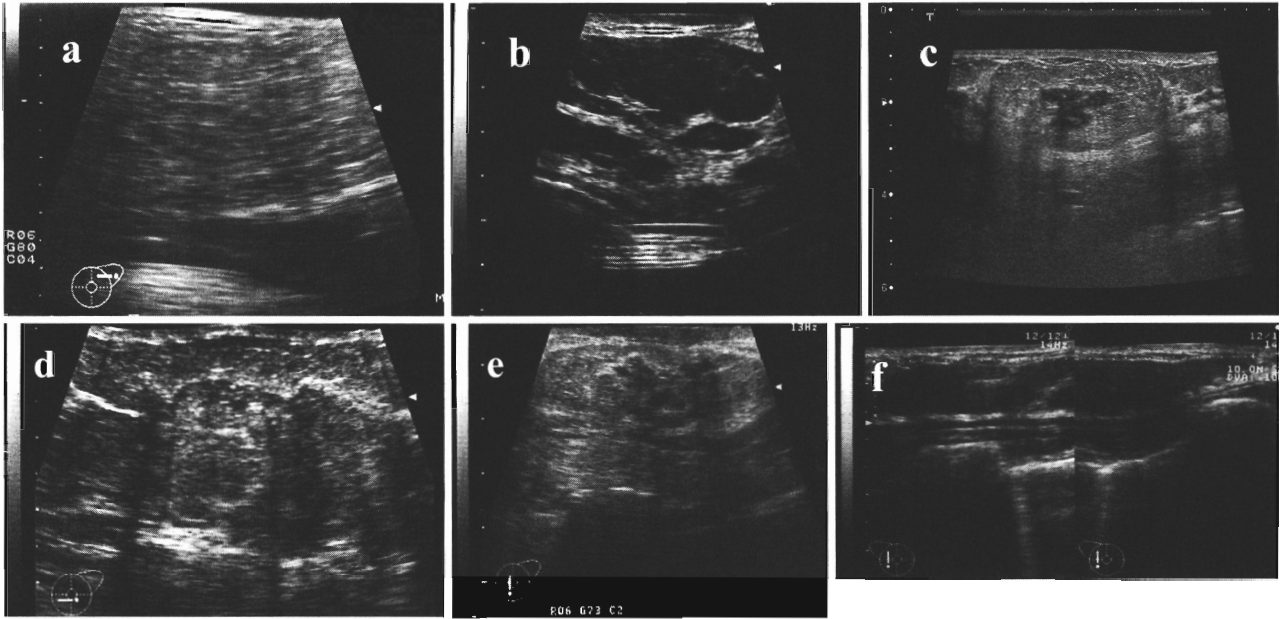


Fig. 2 Mammary ultrasound findings

- a: case a. An oval tumor mass was observed in the left AC region having a somewhat poorly defined border and heterogeneous internal echogenicity.
- b: case b. A shallow spherical tumor mass containing with high and low internal echogenicity was observed in the left ABE region having a well-defined border.
- c: case c. A shallow spherical tumor mass containing with internal marbled echogenicity was observed in the right BD region.
- d: case d. A segmented tumor mass having high and low echogenicity was observed in the left B region.
- e: case e. A shallow spherical tumor mass containing with internal marbled echogenicity was observed in the left AB region.
- f: case f. A shallow spherical tumor mass containing with high and low internal echogenicity was observed in the right BD region having a well-defined border.

resulting in a long duration of symptoms. The image findings depict a tumor mass with circumscribed margins having a well-defined border in all cases, so that it is difficult to distinguish the tumor mass from fibroadenoma^{(3)–(5)}. A radiolucent zone is observed in MMG findings, while breast US findings consist of an oval or segmented shape with a well-defined border, while the internal heterogeneous echogenicity reveals various findings, including a uniform, tachetic, linear or marbled structure. In an MRI, the tumor mass is depicted as having a well-defined border and circumscribed margins, T1-enhanced images of the interior reveals satisfactory distinction with mammary gland tissue with fat components demonstrating well-defined, high signals on T1-weighted imaging. MMG findings show a tumor mass shadowing of localized spherical or oval shape having a radiolucent zone in the surrounding

area. At that time, the cases with much fat tissue in the constituent components shows radiolucent tumor mass shading, and the cases with much gland tissue show dark shading. Similarly, in US findings, high and low echogenicity are internally mixed, which indicates that the internal fat tissue and gland tissue or fibrous tissue are complicated. Hamartoma exhibits peculiar findings because both fat tissue and gland tissue are mixed within the well-circumscribed tumor mass. The radiolucent zone of the tumor mass is peculiar in an MMG and the stripe marble pattern (dendriform) is peculiar in US and an MRI. The description rate of MMG was 82% (cases a, b, c, e, f), US was 33% (cases b, e), and the MRI was 100% (cases a, b, e, f). Because fat tissue was described in the clear high signals on T1-weighted imaging in the MRI, the internal mixture pattern was well described with the contrast with

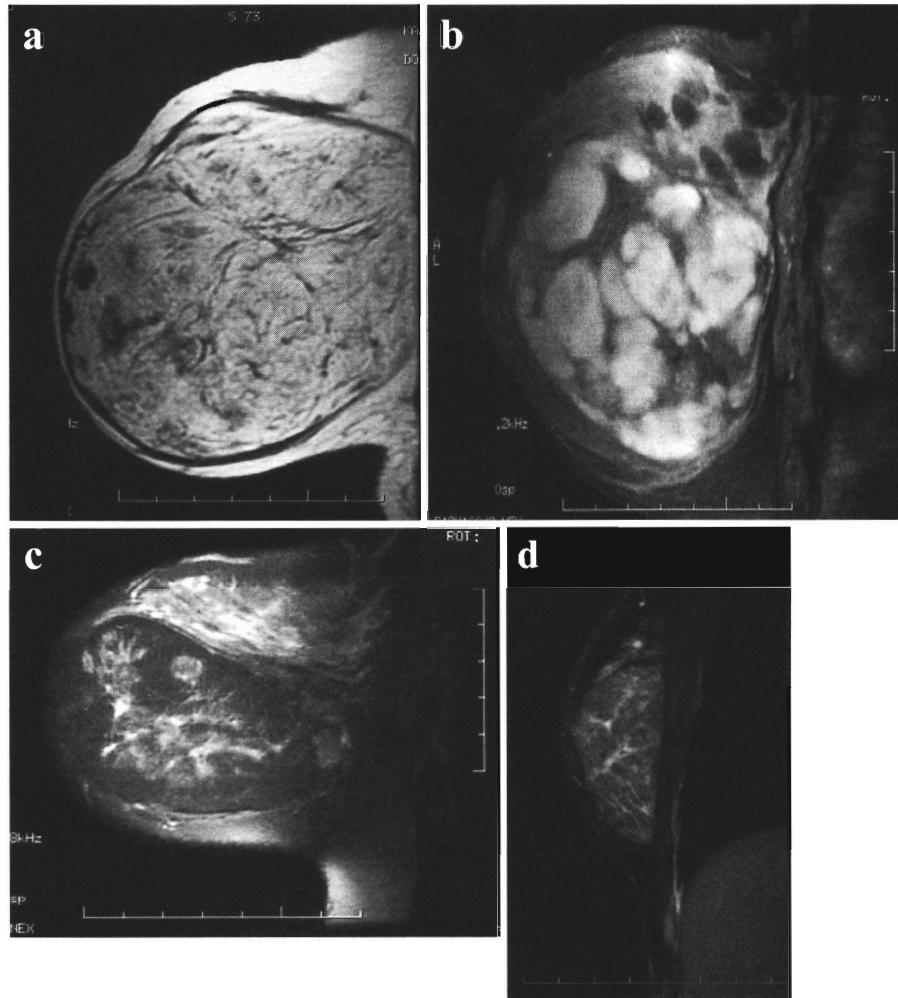


Fig. 3 Mammary MRI findings

- a: case a. A tumor mass having an internal heterogeneous intensity with dendriform and well-defined border was observed in the left breast, and appeared to exclude the normal mammary gland.
- b: case b. A tumor mass having an internal high-intensity with nodular and exhibiting a well-defined border was observed in the left breast, and appeared to exclude the normal mammary gland.
- c: case e. A tumor mass having an internal heterogeneous intensity with dendriform and exhibiting a well-defined border was observed in the left breast, and appeared to exclude the normal mammary gland.
- d: case f. A tumor mass having an internal heterogeneous intensity with dendriform and exhibiting a well-defined border was observed in the right breast, and appeared to exclude the normal mammary gland.

low-signal gland tissue most sharply shown. As the detection rate is 100% even for the fat element in a little tumor mass, the sensitivity is very high. Therefore, this inspection is useful for cases very suggestive of hamartoma.

Cowden disease, a genetic disease, exhibits systemic multiple hamartoma lesion derived from all 3 germ layers, and is complicated by significant rates of malignant tumors such as digestive tract poly-

posis. Our patients did not include cases with this disease. Age distribution for hamartoma in this hospital peaked in premenopausal years, and the incidence is higher than fibroadenoma and lower than breast cancer, but no peculiarities were found (mean age 45.7). Although hamartoma is a benign lesion, since enlargement cannot be predicted even in the case of small tumors, it is difficult to not consider it for surgical removal. However, basic moni-

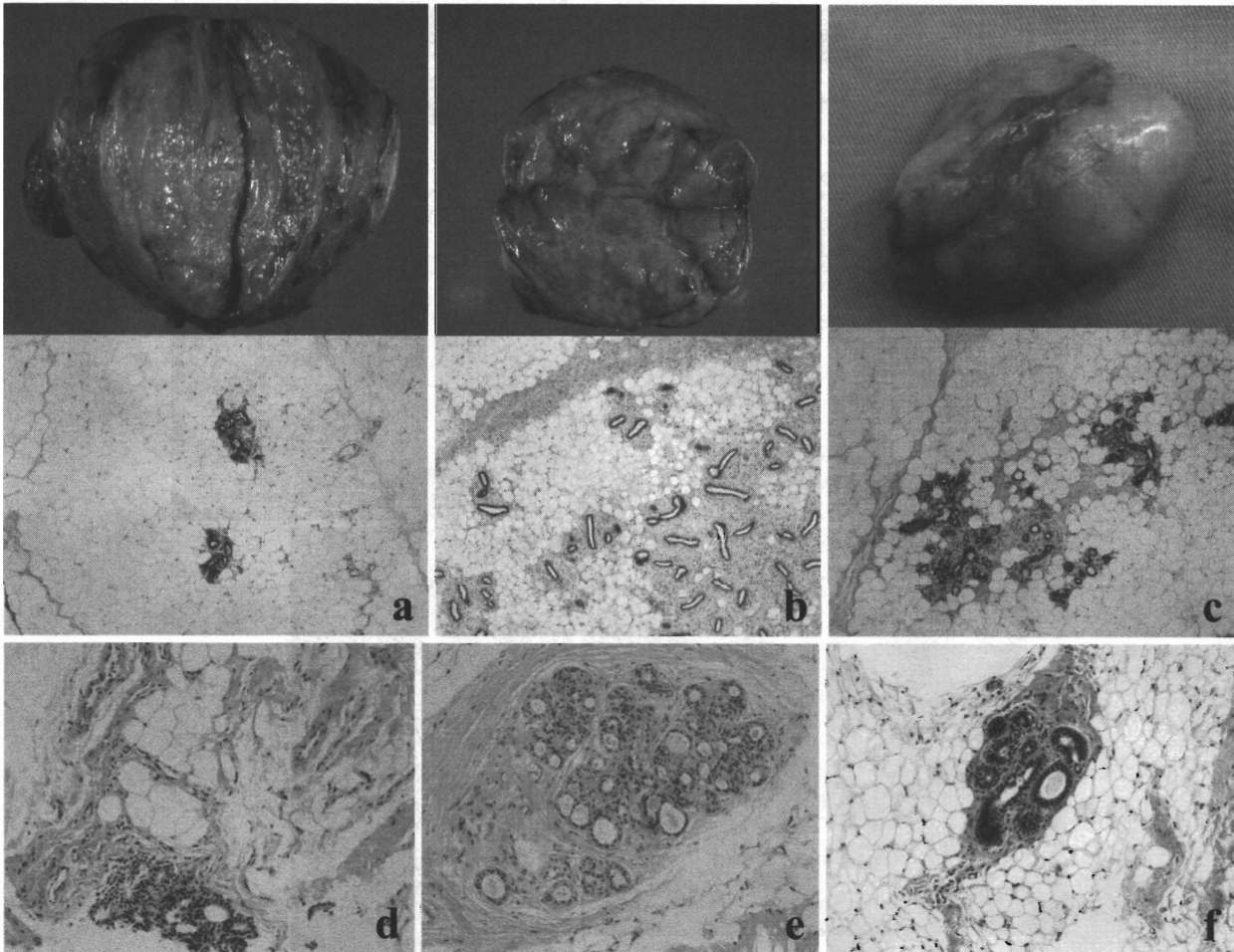


Fig. 4 Macroscopic findings and Pathologic findings of the mammary hamartoma

- a: case a. Lipomatous hamartoma with a capsule, and the cross-section exhibited a partial grayish-white color within yellowish tissue thought to be fat tissue (H-E, $\times 100$)
- b: case b. Fibroadenomatous hamartoma with a capsule, and the cross-section exhibited a fibroadenomatous nodular shape, and had a partial tachetic yellow area (H-E, $\times 100$)
- c: case c. Lipomatous hamartoma with a capsule, and the cross-section exhibited a partial grayish-white color within yellowish tissue thought to be fat tissue (H-E, $\times 100$).
- d: case d. Fibromatous hamartoma. Presence of both fat tissue and mammary gland tissue (H-E, $\times 200$).
- e: case e. Lipomatous hamartoma. Presence of both fat tissue and mammary gland tissue (H-E, $\times 200$).
- f: case f. Lipomatous hamartoma. Presence of both fat tissue and mammary gland tissue (H-E, $\times 200$).

toring of the course of the tumor mass is considered to be adequate with respect to treatment. There are many cases in which the tumor mass reaches a size of 5 cm or more, and in the case of a tendency for the tumor mass to become enlarged, it is necessary to consider surgical excision. In addition, since the tumor may also occasionally be present together with a malignant tumor, for cases in which even some malignant findings are observed, the tumor should be excised even if there are findings characteristic of hamartoma. The use of a proper

procedure enables the tumor to be excised without leaving any disfiguration, thereby making it possible to prevent deformation of the developing breast^{(6)~(8)}. Being familiar with the potential for hamartoma to be found among benign mammary tumors during the course of routine examinations is believed to enable an accurate diagnosis relating to mammary hamartoma, and to lead to appropriate treatment based on its characteristic image findings.

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乳腺過誤腫の6例の検討

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乳腺過誤腫は脂肪成分、乳腺、結合組織から構成され腫瘤を形成し、非腫瘍性の病変であるが腫瘍様病変に分類される。マンモグラフィ検査では radiolucent zone をもつ限局性腫瘤陰影を示し、超音波検査では線維腺腫に似た像を示すが、内部エコーは低エコーと高エコーが混在することが特徴的である。6例のうち radiolucent zone を示したのは5例（82%）、内部エコーが混在していたのは6例（100%）であった。内部エコーが混在する症例のうち過誤腫に特徴的なマーブル模様を呈していたのは2例（33%）であった。MRI 検査では T1 強調単純画像で脂肪は高信号に描出されるために、低信号の腺組織とのコントラストがマンモグラフィ検査や超音波検査に比べ内部構造を明瞭に描出する。MRI 検査を施行した4例では4例（100%）いずれも境界明瞭であり、脂肪組織と腺組織の内部構造のコントラストが明瞭であった。坂元らの組織学的分類から検討すると4例が脂肪腫様過誤腫、2例が線維腺腫様過誤腫であった。良性であるが時に巨大化や乳腺過誤腫内部に潜在した乳癌の報告もあり、特徴的な画像を示すことを熟知していれば診断および適切な治療の選択が容易である。