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CASE REPORT

LEIOMYOADENOMA (MUSCULAR HAMARTOMA) OF THE BREAST

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Abstract

A case of leiomyoadenoma (muscular hamartoma) of the breast is reported here. Only three other cases were found in the literature. The literature is also reviewed on the benign mammary tumors with a mixture of glandular and muscular component.

INTRODUCTION

Abundant smooth muscle elements are rarely recognized in benign mammary tumors. Pure leiomyoma of the breast and nipple[1,2,3], fibroadenoma[4,5], and tumors designated as muscular hamartoma by Davies and Riddell[6] are among these. Recently, we have encountered a breast lesion which we considered to be an example of muscular hamartoma and called leiomyoadenoma descriptively. Only three such cases have been reported previously. The rarity of this tumor prompted us to report our case, and review the literature on benign tumors with a mixture of glandular and muscular components.

CASE REPORT

A 40 year-old female (Gravida 5, Para 2) noticed a right breast mass about a year ago and then left breast masses six months ago. They were asymptomatic. On physical examination, right breast mass and three of the left were found cystic. One of the left breast masses was, however, rather firm and solid, measuring 1 cm in diameter. It was present apparently a few centimeter apart from the lateral edge of the areola. Multicystic lesions were aspirated and the last tumor was removed by local excision.

PATHOLOGICAL FINDINGS

An excised mass was oval, well-demarkated, solid tumor, which measured 1.5 cm in the greatest dimension. It was firm in consistency. Microscopically

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it contained numerous medium-sized ducts, ductules and acini. Lobular architecture was mostly indistinct with abundant muscular investment, but focally well-preserved (Fig. 1). Smooth muscle cells tended to focally gather forming compact bundles especially in areas adjacent to ducts and ductules (Fig. 2). They also streamed singly into the lobules and between acini. Those cells were identified as smooth muscle cells by the presence of longitudinal filaments in PTAH and Azan-Mallory stained sections. A few arterioles with muscular wall were scattered in the stroma, but there were no transitions between these blood vessels and muscle bundles mentioned earlier.

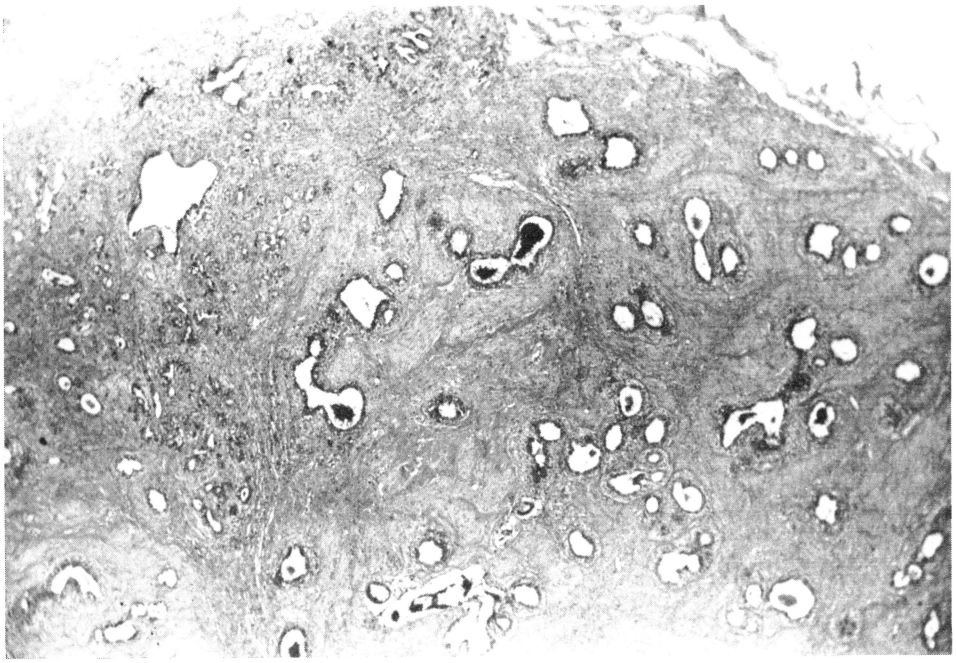

Fig. 1. Microscopic appearance of leiomyoadenoma.
Dilated ducts and smooth muscle bundles are intermingled.
A few areas of mammary acini are still preserved.
(H-E, Original magnification × 20)

DISCUSSION

The presence of smooth muscle cells within benign mammary tumors has been rarely reported[1~8]. Pure leiomyomas of the breast[1,2] and nipple[3] are one example of such tumors. Benign tumors with a mixture of epithelial and muscular components have been variously designated ; namely, fibroadenoma with smooth muscle[4,5], adenoleiomyoma[7], and muscular hamartoma[6]. Table 1 summarizes pertinent clinico-pathological findings of those cases appeared in the

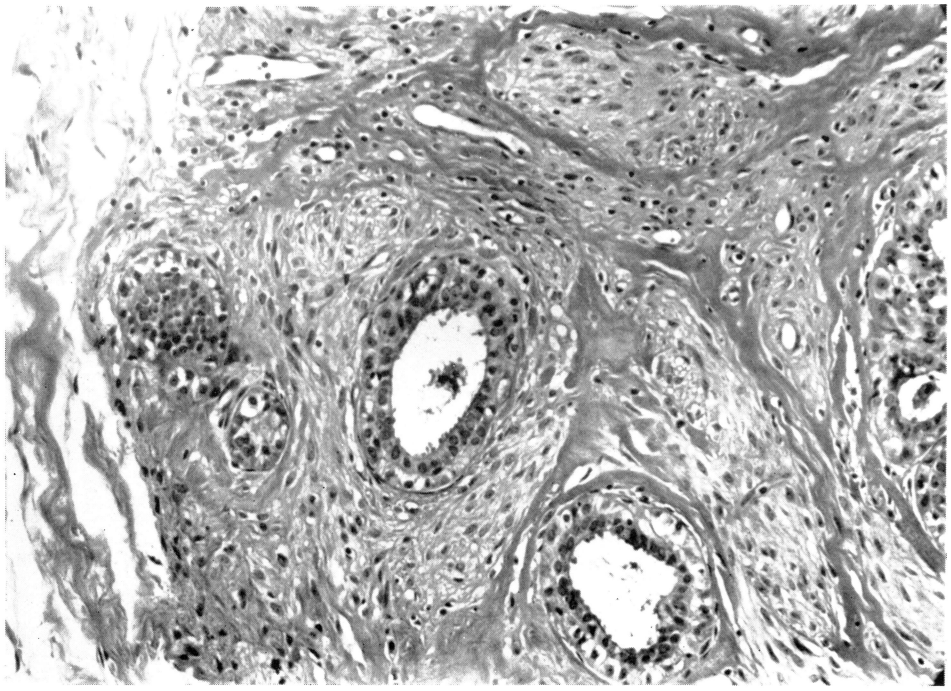

Fig. 2. Higher magnification of Fig. 1.
Note that epithelium-lined ducts are surrounded by bundles of smooth muscle cells.
(H-E Original magnification × 200)

literature. We are not aware of similar case reports in Japanese literature.

As the name implies, fibroadenoma should be composed of epithelial and fibrous tissue elements. The latter element is a loose fibroblastic stroma with abundant mucopolysaccharide in typical cases. There are, of course, marked variations in cellularity. Hyalinization of the stroma is common. Cartilagenous, osseous, or adipose metaplasia of the stroma are rarely seen. In order to diagnose fibroadenoma with smooth muscle, the presence of the areas showing typical appearance of fibroadenoma seems essential. Cases reported by Mackenzie[4], and Goodman and Taxy[5] fulfilled this criteria, thereby designated properly. From the lack of such appearance as well as the coexistence of adipose tissue in one of their cases, Davies and Riddell[6] named their cases as muscular hamartoma. Haagensen[7] described a similar case without adipose element under the term of adenoleiomyoma. In our opinion those three cases were probably same in nature.

The term hamartoma was coined by Albrecht[9] in 1904 to describe tumors resulting from a localized error in development of a normal component or components of an organ. A hamartoma is usually confined to a limited

TABLE 1

Cumulated cases of benign mammary tumors with a mixture of glandular and muscular components

Reference	Age	Symptom	Duration	Location and size	Histology	Term applied
1. Davies & Riddell (6)	44	painless swelling	3 mos.	Right ; upper outer quadrant 7×5×3 cm	Ducts ; ductules and lobules. Muscle bundles.	Muscular hamartoma
2. Davies & Riddell (6)	48	painful, mobile swelling	2 wks.	Right ; immediately lateral to the areola 3.5×3×2.5 cm	Lobules and ducts. Bands of muscle cells. Islands of fat cells.	Muscular hamartoma
3. Haagensen (7)	40	mass	1 mos.	Left ; upper outer quadrant 3 cm	Groups of small ducts or acini. Smooth muscle cells with the appearance of leiomyoma.	Adenoleiomyoma
4. present case	40	mass	6 mos.	Left ; a few cm lateral to the edge of areola 1.5 cm	Ducts, ductules and acini. Muscle bundles around ducts.	Leiomyoadenoma
5. Mackenzie (4)	60	slightly tender lump	6 wks.	Left ; 2.5 cm	Ducts. Stroma with smooth muscle cells and occasional myxomatous change.	Fibroadenoma with smooth muscle
6. Goodman & Taxy (5)	39	nontender mass	ND	Left ; adjacent to the lateral edge of the areola ; 4×5 cm	Duct proliferation. Stroma which was fibrous and collagenous with foci of large interwining bundles of smooth muscle cells.	Fibroadenoma with prominent smooth muscle
7. Goodman & Taxy (5)	51	mass	18 mos.	Left ; upper inner quadrant at the border of areola 3×4 cm		Fibroadenoma with prominent smooth muscle
8. Cheatle and Cutter cited in (4 & 6)	ND	ND	ND	ND	ND	Intracanalicular fibroadenoma with smooth muscle
9. Azzopardi (8)	ND	ND	ND	ND	ND	Smooth muscle in fibroadenoma
10. Azzopardi (8)	ND	ND	ND	ND	ND	Smooth muscle in fibroadenoma

ND : no description

anatomical field and various names are applied for the same lesions depending upon the proportion of its constituents. For instance, synonyms for the hamartoma of the lung include adenochondroma, chondromatous hamartoma, fibroadenoma of the lung, pulmonary lipochondroadenoma and so forth.

Muscle elements in the fibroadenoma may result from metaplasia of the connective tissue or from myoid differentiation of the myoepithelial cells[8]. In such cases, muscle cells may assume random orientation without bundle formation. Our findings (1) that typical appearance of fibroadenoma was not present, (2) that the acinar or lobular structures were still preserved and (3) that smooth muscle cells were focally aggregated to form compact bundles around ducts are suggestive of hamartomatous nature of this tumor. Like other hamartomas, the descriptive term leiomyoadenoma or adenoleiomyoma would be most appropriate in our case, and we favor the former because of the abundance of epithelial elements.

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