Muscular Hamartoma of the Breast : A Rare Breast Lesion Containing Smooth Muscle

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Astract

Hamartoma of the breast is an uncommon entity, usually presenting as a well-demarcated breast mass. Microscopically, the lesion is composed of mammary glandular component, fibrous stroma, adipose tissue, and smooth muscle in variable proportions. Among the variants of breast hamartoma, muscular hamartoma is rare. This lesion should be differentiated from other breast tumors that contain smooth muscle element.

We report a breast lesion of a 36-year-old woman diagnosed as a muscular hamartoma in which the muscular component is cellular and some mitotic figures are present. The criteria to distinguish between benign and malignant smooth muscle lesions in the breast, emphasizing mitotic count, are also discussed.

Hamartoma of the breast is characterized by a well-demarcated breast lesion comprising a variable mixture of mammary epithelial element, fat, fibrous tissue, and, occasionally, smooth muscle in variable proportions(1). This benign lesion has been considered to be uncommon, but has become more frequently encountered with the increase of mammographic screening(2). Muscular hamartoma represents an unusual variant of breast hamartoma that contains prominent stromal components of smooth muscle(3). It is important for pathologists to be aware of this entity since it seems to be underdiagnosed, even in the case in which a breast mass is clearly demonstrated by mammography(4). The confusing points concerning hamartoma of the breast, muscular variant, which need to be clarified and discussed include the following : 1) the criteria to diagnose and to distinguish from other lesions which can also reveal the presence of muscular components, and 2) the criteria to distinguish between benign and malignant lesions.

CASE REPORT

A 36-year-old woman presented with a painless left breast mass. Four years previously, the lesion was detected as a 1 cm circumscribed

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soft mass by the physician. The lesion remained unchanged until last year, when it became progressively enlarged. On examination, a 7 cm circumscribed, firm, and mobile mass was palpated at the inner lower quadrant. Surgical removal of the entire breast mass and adjacent breast tissue was performed and the specimen was sent for histopathologic examination at Maharaj Nakorn Chiang Mai Hospital.

Clinical information revealed that the patient had two full term pregnancies and had regularly taken oral contraceptive drugs for about ten years. Her obstetric and menstrual histories were not remarkable.

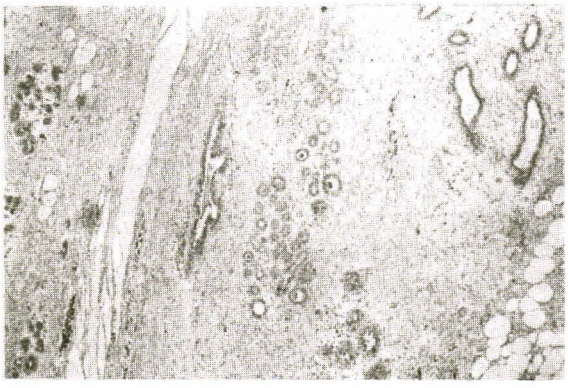

Fig. 1. Mass (right of field) composed of mammary ducts and lobules, fibrous stoma, adipose tissue, and smooth muscle separated from adjacent tissue by a sharp interface. (H&E,x 40)

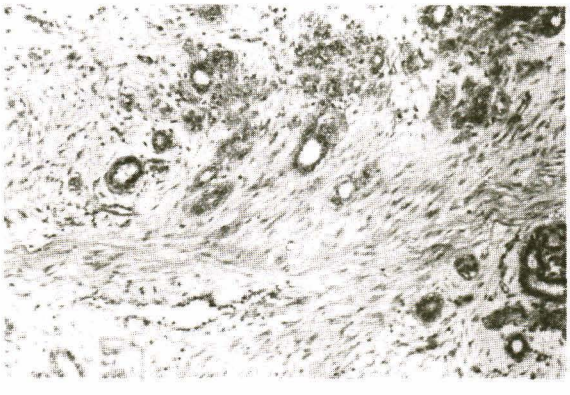

Fig. 2. Smooth muscle bundles closely related to foci of sclerosing adenosis with myoepithelial proliferation. (H&E,x 100)

Fig. 3. Compact bundles of smooth muscle cells. (H&E,x 100) Inset, uniform smooth muscle cells showing eosinophilic cytoplasm and blunt-ended nuclei. (H&E,x 400)

On gross examination, the lesion was a well-circumscribed ovoid mass measuring 7 x 5 x 5 cm. The cut surface was gray-white and firm. A few small cysts filled with clear fluid, measuring 0.1 up to 0.5 cm, were observed. Histologic examination revealed an unencapsulated mass separated from adjacent breast tissue by a sharp interface (Fig. 1). The lesion contained numerous irregularly-arranged mammary ducts and lobules with loose paucicellular stroma sparsely infiltrated by lymphocytes and plasma cells. Foci of sclerosing adenosis with proliferation of myoepithelial cells were observed in some areas (Fig. 2). Cystic dilatation of the mammary ducts with apocrine metaplasia was noted. Closely surrounding the glandular component were bundles of uniform elongated spindle-shaped cells with eosinophilic cytoplasm (Fig. 3). Their nuclei were uniform and vesicular with blunt ends. Focal areas of mature fat tissue admixed with fibrous stroma and eosinophilic spindle cells were observed (Fig. 1). Mitotic figures without atypical form were occasionally present in the glandular element and the fibrous stroma, whereas, rare mitotic figures (approximately 1 in 50 high power fields) were seen in the areas of eosinophilic spindle cells. There was no stromal hypercellular area, nuclear pleomorphism, or abnormal mitosis detected.

Further studies to identify the specific cell type of those eosinophilic spindle cells were performed. By trichrome stain, the cytoplasm showed

intense reddish staining. Immunohistochemical stains revealed positive intracytoplasmic staining with anti-smooth muscle actin and anti-desmin antibodies. Negative result was obtained by using anti S-100 antibody. These results supported the smooth muscle nature of the eosinophilic spindle cells detected by H&E staining. Ultrastructural study was performed on the tissue that was already fixed in formalin. The spindle cells revealed numerous intracytoplasmic myofilaments and scattered fusiform dense bodies ; however, some other ultrastructural details were rather poorly preserved due to initially improper fixation.

The postoperative course was uneventful. There was no recurrence within 6 months postoperatively.

DISCUSSION

Hamartoma of the breast was first defined by Arrigoni et al[5] as a well-demarcated encapsulated breast tumor composed of mammary glandular tissue with prominent lobular arrangement, fibrous stroma, and fat in variable proportion. This entity is now well recognized in the radiologic literature[4]. Macroscopically, the lesions are usually well-defined, round to oval or lens-shaped masses. On histologic examination, they are sharply circumscribed with or without capsule[2,4]. There is a spectrum of change ranging from normal mammary tissue to the full gamut of change in benign breast disease[1]. It was suggested by Jones et al[6] that variable histologic patterns of breast hamartoma may represent the results of physiologic changes in the mammary gland. Practically, diagnosis of breast hamartoma is based on exclusion of other well characterized benign mammary tumors[5]. However, the term breast hamartoma was probably not uniformly used among the investigators since some benign breast lesions such as adenolipoma have been accepted as breast hamartoma by some authors but not by others[7].

In 1973, Davies & Riddell[8] described 2 cases of "muscular hamartoma", which later on were accepted as distinctive and extremely rare variants of mammary hamartoma[9]. The origin of smooth muscle in muscular hamartoma is uncertain but is attributable to myoepithelium, muscle in the walls of local blood vessels, and undifferentiated mammary mesenchyme[8]. To our knowledge, approximately eleven cases of muscular hamartoma have been reported in the literature[1,3,8,10-13]. Clinical data of these patients are summarized in Table 1. The patients' ages ranged from 34 to 61 years (mean 44.3 years). Most patients presented a breast mass with a duration ranging from 2 weeks to 4 years (mean 18.8 months). The lesions varied in size from 2.5 cm to 8 cm (mean 5.0 cm). Compared to the usual mammary hamartoma, these values were not significantly different[4]. Concerning the description of muscular hamartoma mentioned, we believe that the lesion of our patient reasonably deserves the description of muscular hamartoma. Numerous eosinophilic spindle cells found were morphologically characteristic of smooth muscle. This was supported by immunohistochemical studies as mentioned earlier. Regard-

Table 1. Summary of the clinical information of the reported cases of mammary muscular hamartoma.

Author(s)	No. of case(s)	Age (Years)	Side	Duration of symptom	Size (cm)	Operation	Follow-up (months)	Result
Davies&Riddell[8]	2	44	Rt.	3 Mo	7	Excision	40	NED
		48	Rt.	2 Wk	3.5	Excision	NI	NI
Huntrakoon&Lin[10]	1	34	Lt.	0	8	Excision	NI	NI
Bussolati et al[11]	1	44	Rt.	0	5.5	Enucleation	6	NED
Daroca et al[12]	3	39	Rt.	0	3	Excision	13	NED
		61	Rt.	1 Mo	3.5	Excision	5	NED
		38	Lt.	1 Y	2.5	Excision	NI	NI
Shepstone et al[13]	1	52	Rt.	4 Y	7	Enucleation	6	NED
Fiirgaard&Kristensen[3]	1	47	Rt.	NI	3.5	Enucleation	NI	NI
Fisher et al[1]	2	NI	NI	NI	NI	NI	NI	NI
Present report	1	36	Lt.	4 Y	7	Excision	6	NED

NED : No evidence of disease ; NI : No information ; 0 : Asymptomatic mass detected during routine physical examination

ing ultrastructural studies in muscular hamartomas, it has been reported that the spindle cells had characteristic features of smooth muscle[10,12,13]. The typical findings included numerous intracytoplasmic actin microfilaments with interspersed fusiform dense bodies, plasmalemmal attachment plaques, surface pinocytic vesicles, and external lamina[10]. Although the ultrastructural details were not completely preserved in our case, presence of numerous intracytoplasmic myofilaments and scattered fusiform dense bodies were consistent with smooth muscle nature of these spindle cells.

To distinguish muscular hamartoma from other smooth muscle-laden lesions, including fibroadenoma and phyllodes tumor, and smooth muscle tumor, the criteria used are discussed as the following. In fibroadenoma, besides smooth muscle component, the stroma is characteristic of typical fibroadenoma with foci of myxomatous change[14]. Moreover, fibroadenoma with smooth muscle component reveals neither preserving of mammary lobular structures nor adipose tissue[6]. In phyllodes tumor, a hypercellular stroma with foci of irregular intracanalicular growth and presence of mitoses are characteristic[4]. Concerning smooth muscle tumor of the breast, admixture of disorganized glandular structures and smooth muscle in a well-circumscribed lesion and presence of fat and fibrous tissue are not the consistent features[15].

In the present lesion, mitotic figures were occasionally encountered in the fibrous stroma but very rare in the smooth muscle element. In breast hamartoma, mitotic figures were reported to be absent or extremely rare by some authors[4]. Concerning muscular hamartomas, mitotic figure in muscular component was absent when it was mentioned[8,10,12,13]. In addition, neither recurrence nor metastasis was reported in the follow-up duration of 5 to 40 months following surgery. Therefore, muscular hamartoma seems to have a benign course. However, in the presence of mitotic figures, it is still important to distinguish between malignant and benign lesions. Regarding smooth muscle tumor of the breast, the tumor with 3 or more mitotic figures/10 HPF was considered leiomyosarcoma, while benign tumor should show no mitosis[16]. In spite of this suggestion, occasional mitotic figures were reported in fibroadenoma with smooth muscle[14] and rare mitoses in some leiomyomas of the breast as well[15]. Although the definite histologic criteria to determine the malignant potential of smooth muscle tumor of the breast has not been settled[15], in our opinion, mitotic count in smooth muscle lesion of the breast should be used as a guideline, in association with other histologic parameters, to predict the outcome for an individual patient.

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ฮามาร์โตมากล้ามเนื้อของเต้านม : รอยโรคที่พบได้ยากของเต้านมซึ่งประกอบด้วยกล้ามเนื้อเรียบ

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Hamartoma ของเต้านมเป็นภาวะที่พบไม่บ่อย ส่วนใหญ่พบรอยโรคเป็นก้อนที่เต้านมซึ่งมีขอบเขตชัดเจน โดยมีลักษณะทางกล้องจุลทรรศน์ที่ประกอบด้วย mammary gland, fibrous stroma, และ เนื้อเยื่อไขมัน ในสัดส่วนที่แตกต่างกัน muscular hamartoma จัดเป็น variant หนึ่งของ hamartoma ของเต้านม ซึ่งพบได้ยาก การวินิจฉัยรอยโรคชนิดนี้จำเป็นต้องวินิจฉัยแยกโรคจากก้อนที่เต้านมชนิดอื่นที่มี smooth muscle เป็นส่วนประกอบ

ผู้รายงานได้นำเสนอก้อนที่เต้านมของผู้ป่วยหญิงอายุ 36 ปีหนึ่งราย ที่ได้รับการวินิจฉัยเป็น muscular hamartoma โดยส่วนของ smooth muscle นั้นมีจำนวนเซลล์มาก และมี mitosis จำนวนเล็กน้อย รวมทั้งได้อภิปรายเกี่ยวกับเกณฑ์ในการวินิจฉัยแยกระหว่างรอยโรคซึ่งประกอบด้วย smooth muscle ที่เป็น benign กับ malignant โดยอาศัยการนับจำนวน mitosis

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ERRATUM

Mo-Suwan L et al. Calcium and phosphate solubility curves for parenteral nutrient solutions containing Vaminolact® 1997; 80: page 517.

* $TA = (V_B - N_B \times 1000) / V_S$

should be :

* $TA = (V_B \times N_B \times 1000) / V_S$