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AGGRESSIVE FIBROMATOSIS INFILTRATING BREAST REPORT OF CASE AND REVIEW OF LITERATURE

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INDEXING WORDS

aggressive fibromatosis; breast; carcinoma; cytology

SYNOPSIS

We report on a 23 year-old female with heterochronic and multiple aggressive fibromatosis on the upper and lower quadrant of the left breast 3 years apart. The tumor was situated on the fascia of the greater pectoral muscle and growing into the mammary tissue, according to ultrasonography. Clinically it was hardly distinguishable from breast cancer, thus a wide resection includ-

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ing sufficient peripheral tissue was performed; up to date no recurrence or remote metastasis have been observed. Histologically long spindle fibroblasts grew with abundant collagen fibers in the interstitial tissue, but cellular density was low and no mitosis was observed. As severe infiltration into surrounding adipose tissue, muscle and mammary gland was indicated, it was diagnosed as aggressive fibromatosis. To distinguish it from breast cancer, definite preoperative diagnosis by histological study is required to avoid unnecessary radical mastectomy.

INTRODUCTION

Aggressive fibromatosis is a pseudotumoral lesion also called desmoid tumor, a non-malignant fibroblastic proliferation observed on the abdominal wall of multipara, producing an intense invasion of the peripheral tissue.[6, 7] It occasionally causes local recurrence, but remote metastasis is noted quite rarely. Meanwhile, lesions of fibroblastic proliferation on other sites than the abdominal wall, namely on the upper arm, shoulder,[7] neck, axilla, hip, etc., are well known as extra-abdominal desmoid tumors with the same histological picture. They are also reported on the thoracic wall, but on the breast are quite rare.[8] The authors experienced a case of aggressive fibromatosis with multiple onsets 3 years apart on the left breast of a 23 year-old woman. This is discussed and the literature.

CASE REPORT

The patient is a 23 year-old unmarried woman. In about May, 1984, she felt a lump on the lower outer quadrant of the left breast, but no treatment was made. On November 21, she visited the surgical clinic of Yashiro Municipal Hospital because the lump was larger and tender and she was admitted. Meanwhile, 3 years ago she had had a tumor on the upper outer quadrant of the left breast, which was extirpated at Nishiwaki Municipal Hospital being diagnosed as fibro-adenoma. Upon examination her general state was found favorable but a hard platelike mass of walnut size was palpable on the lower outer quadrant of the left breast. The mass was tender to pressure. A shallow dent was noted on the skin

thereabout. There was no adhesion with the skin but with the underlining tissue. Furthermore, on the upper outer quadrant of the left breast a scar of the previous operation was noted, which however was not related to the mass found this time. There was no history of contusion, nor surgical invasion. Abnormal secretion from the nipple was not observed.

Upon ultrasonography of the left breast, a mass with low echo area of 4 x 1cm in size growing in the breast riding over the greater pectoral muscle was noted on the echogram together with increased echo at the posterior wall and uneven echo-image inside the mass (Fig. 1). On the breast X-ray film, a shadow mass was unclear without showing calcification.

Suspecting breast cancer, an aspiration biopsy was conducted on the mass. Cytologically, a few spindle cells with egg-shaped nucleus were noted. Some epithelial cells were not found atypism, therefore, it was suspected to be fibroadenoma or fibroma (Fig. 2). The clinical test showed nothing abnormal in the peripheral blood, liver functions, urinalysis or serological analysis. Upon barium enema, no polyposis was found in the large intestine.

Suspecting breast cancer, a wide resection of the mass was carried out on November 29th. Postoperative course was favorable, and up to date no recurrence nor remote metastasis has been observed.

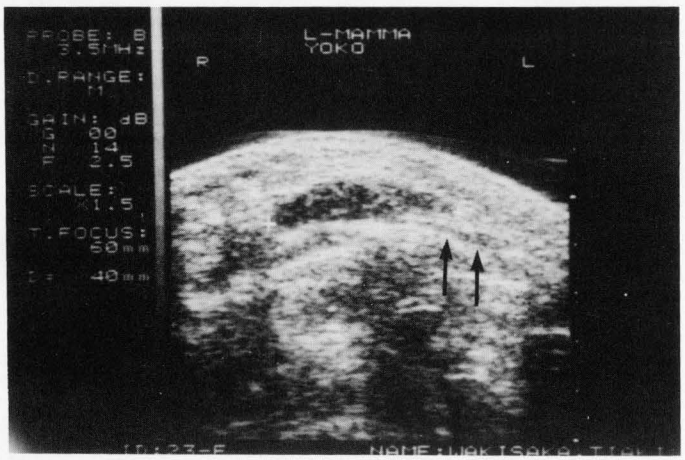

Fig. 1 Ultrasonography of the left breast tumor.
A low echo area of 4 x 1cm deep in mammary gland growing over the pectoral muscle (arrow) is observed.
Uneven image of internal echo can be seen.

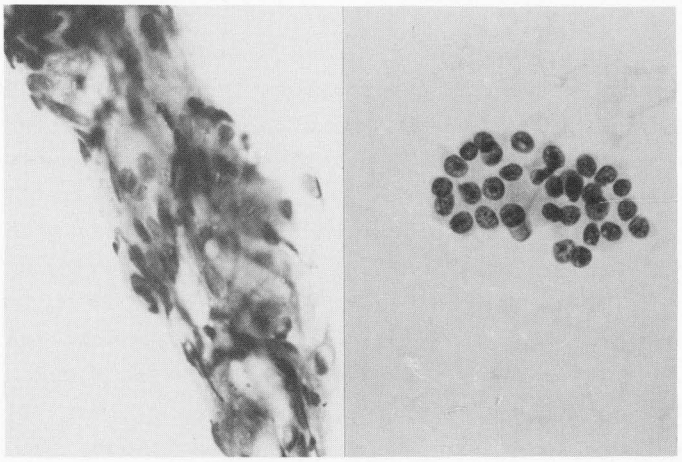

Fig. 2 Cytological findings from the left breast tumor.
Left: A fusiform nucleus with small nucleolus is found in the plump spindle cytoplasm, however, irregularity in size, increase of nuclear chromatin and atypical mitosis is not observed in these cells. Papanicolaou's stain, x 30.
Right: Another cell nest consists of round epithelial cells with small round nuclei, which have no atypism. Papanicolaou's stain, x 30.

PATHOLOGICAL FINDINGS

Macroscopic findings: The mass extirpated was located deep in the mammary gland at the lower outer quadrant of the left breast, in a flat plate form of 4 x 4 x 1.5cm, hard with no apparent capsule, a yellowish white fibrous mass with no cyst formation, necrosis nor bleeding (Fig. 3).

Microscopic findings: The tumor consisted of spindle cells with eosinophilic fibrous stroma, showing infiltrative growth into the surrounding tissue (Fig. 4; left). It had no capsule. Furthermore, it also infiltrated into muscles, atrophied muscular bundles remaining in the mass (Fig. 4; right). In the part grown upward, it was growing and enveloping the mammary tissue (Fig. 5; left), thus the enveloped mammary tissue was atrophied. Part of the mammary duct incorporated in the mass (Fig. 5; right), showed

a double-layer growth of the epithelium suggesting apocrine metaplasia. Around these cells meyoepithels with pale cytoplasm were observed. However, no transformation to the surrounding spindle cells was noted. Growing cells had spindle and slender cytoplasm with fusiform nucleus, poor in chromatin, and a few small nucleoli. Among these cells abundant collagen fibers were noted, but inflammatory cell infiltration was scarcely to be seen (Fig. 6). No mitosis was noted, and the density of proliferating cells was low. In cytoplasm, no periodic acid-Schiff (PAS) positive material nor phosphotangstic acid hematoxylin (PTAH) positive microfibrils were observed.

From the above the mass was diagnosed as aggressive fibromatosis involving the mammary gland.

The mass on the upper outer quadrant of the left breast extirpated 3 years ago indicated the same histopathological findings as those noted this time (Fig. 7), showing no growth of the epithelial component; therefore, it was also diagnosed as having been aggressive fibromatosis. Meanwhile, as the mass this time is situated far from that of 3 years ago, the fibromatosis was considered to be multiple and heterochronic.

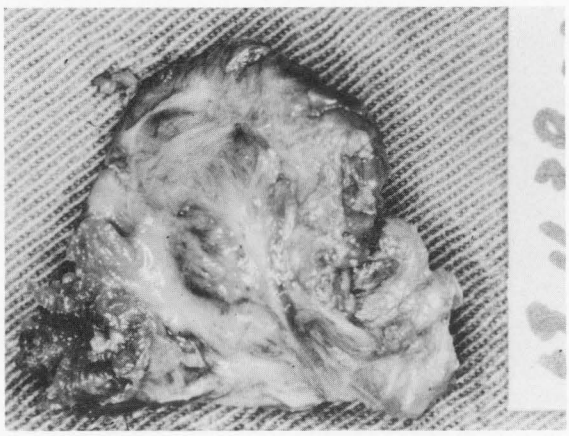

Fig. 3 Macroscopic finding of the extirpated tumor.
Flat plate-like tumor of 4 x 4 x 1.5cm without definite capsule shows yellowish white cut surface and has no bleeding nor necrosis.

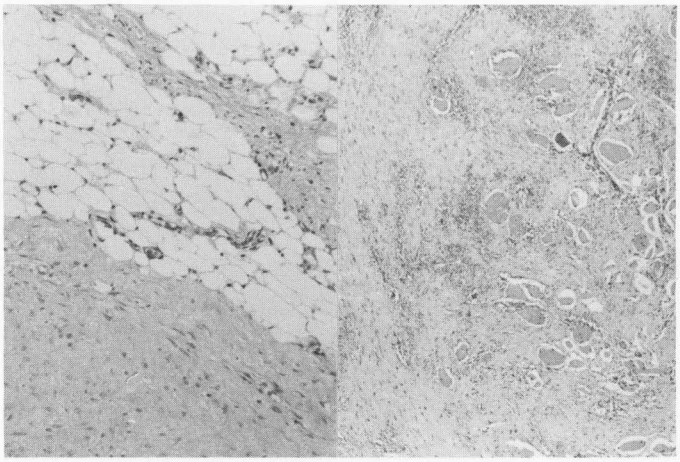

Fig. 4 Histopathological findings of the extirpated tumor.
Left: It shows infiltrative growth into the surrounding adipose tissue. H.E.stain. x 1.5.
Right: Striated muscle remains scattered around the tumor due to infiltrative growth by the tumor. H.E. stain, x 1.5.

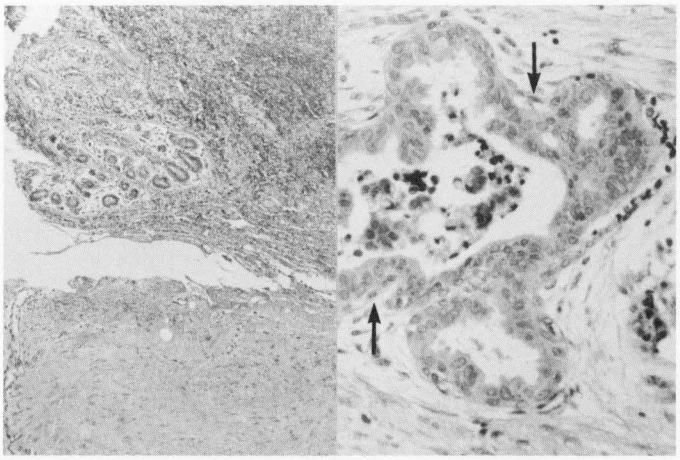

Fig. 5 Histopathological findings of the extirpated tumor.
Left: Mammary duct is surrounded by the infiltrating tumor. H.E.stain, x 3.
Right: Epithelium of the remaining mammary duct is double layered, backing of myoepithels with pale cytoplasms (arrow) can be seen. H.E.stain, x 15.

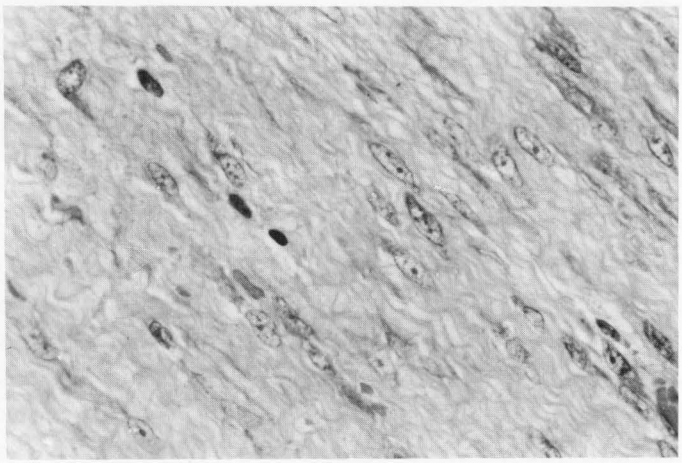

Fig. 6 Histopathological finding of the extirpated tumor. Nucleus of the tumor cell is fusiform or egg-shaped with a small nucleolus, and cytoplasm looks eosinophilic. Among tumor cells abundant collagen fibers are seen. No nuclear mitosis is observed. H.E.stain, x 30.

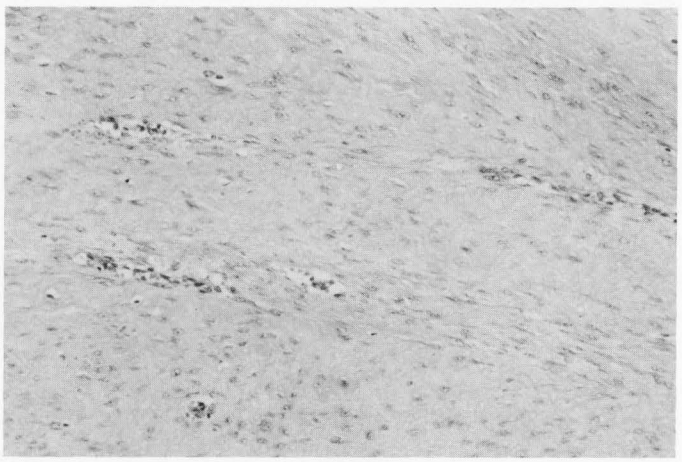

Fig. 7 Histopathological finding of the tumor on the upper outer quadrant of the left breast extirpated 3 years ago. Same as this time, it is a tumor comprising proliferation of spindle cells with no hyperplasia or atypism of mammary epitelial component. H.E.stain, x 15.

DISCUSSION

Desmoid tumor other than on the abdominal wall (aggressive fibromatosis) is known as pseudotumoral fibroblastic proliferation derived from fascia of voluntary muscles other than the abdominal wall and was observed in adults of 20 to 30 years.[6] Occurence in children is also reported. Histologically the difference from abdominal desmoid is not clear. It frequently occurs on the upper arm, lower leg, thigh, and is also reported to be found on the shoulder, neck, axilla, hip, thoracic wall, etc., but rarely on the mammary gland. It is a grayish white pseudotumoral lesion without capsule in gross. Histologically it consists of a mature fibroblastic proliferation showing abundant collagen fiber formation in its stroma. Under electron microscopy some cases are reported to have myofibroblasts mixed in the cells comprising the mass. As to prognosis it is reported that local recurrence is noted at a high ratio, but no remote metastasis is observed. It is said to be treated by wide extirpation involving sufficient surrounding tissue.

Differential diagnosis is necessary from well differentiated fibrosarcoma, tumors originating from smooth muscle, and nodular faciitis. The well differentiated fibrosarcoma is distinguishable as it shows a higher cellular density than aggressive fibromatosis, atypical nuclear mitosis, and causes remote metastasis. It is differentiated from tumors originating from smooth muscle in shape of nucleus, presence of PTAH positive intracytoplasmic microfibril and collagen fiber in its stroma. Nodular fasciitis shows a specific clinical course and mostly degenerates within a week or two spontaneously. Histopathologically, it comprises a fibroblastic proliferation, but has little collagen fiber in its stroma and inflammatory cell infiltration mixed therin.

Furthermore, a complication with multiple polyposis in the large intestine is reported, summarized as Gardner's syndrome; it involves complications with fibroma, osteoma and other soft tissue tumors.

Up to date 39 cases have been reported as aggressive fibromatosis onset on the mammary gland (Table 1).[1, 2, 3, 4, 5, 8, 9, 10, 11, 12, 13, 14, 15, 16, 17, 18, 19, 20] All cases are female and from 18 to 80 years old (mean; 44. 2). The left breast is involved more than the right (11:5). The size of mass ranges from

1.0 to 10.0cm in diameter. Seventeen cases were preoperatively misdiagnosed as carcinoma of the breast. Gardner's syndrome are found in three cases,[11, 19, 20] two of these three being multiple [19, 20] and another case bilateral.[11] Seven cases involve the underlining pectoral muscle and infiltration into the mammary gland,[1, 10, 11, 12, 13, 18] and are hard to differentiate clinically and histologically from that on the mammary gland itself.

The lesion on the left breast of the 23 year-old woman reported in this paper shows intense infiltrative growth into the peripheral adipose tissue and muscles involving mammary tissue in the mass. It leans over the fascia of the greater pectoral muscle, seemingly originating therefrom, but apparently it infiltrated into the mammary tissue and is hardly differentiated from breast cancer by inspection, palpation or ultrasonographic examination. Thus, aspiration biopsy was executed, from the findings of which the tissue was assumed to consist of plump spindle cells originating from mesenchymal tissue. However, quite a few epithelial components were observed, which is a notable finding. In cases when such cytological findings are indicated, the possible presence of aggressive fibromatosis on the mammary gland should also be taken into consideration. Extirpation is conducted rather widely. As no recurrence has been observed up to date, mastectomy should not be done easily.

In this patient the same lesion noted at the upper outer portion of the left breast was extirpated 3 years ago, and revealed the same histopathological findings. However, as there is no continuity between them it is considered to be a heterochronic and multiple occurrence. Meanwhile, in this case polyposis of the large intestine has not been confirmed. Multicentric fibromatosis without Gardner's syndrome is reported in this paper for the first time.

With this case too, aggressive fibromatosis on the mammary gland is hard to differentiate from breast cancer clinically, and requires accurate preoperative diagnosis by means of cytological (aspiration biopsy) and histopathological (open biopsy) techniques. It is considered important to prevent unnecessary radical mastectomy.

Table 1 Reported cases of aggressive fibromatosis of the breast

Author(s) (year)	No. of cases	Age	Location*	Size	Involvement of pectoral muscle
Nichols (1923)	1	36	right,LIQ	5x4x2	+
Adair & Hermann (1946)	1	32	left,UIQ	2	+
Prior & Sisson (1954)	1	27	left		
Simpson et al. (1964)	1	38			
Cammeron & Adair (1965)	2				
Norris & Taylor (1968)	2				
Das Gupta et al. (1969)	3				
Zayid & Dihmis (1969)	1	43	left,upper		
Haggitt & Booth (1970)	1	21	left,UIQ right,UIQ	6x4 3x2	+ +
Schremmer (1971)	1	51	left,UOQ		+
Kalisher et al. (1976)	1	50	left,UOQ	5x3	+
Rosen et al. (1978)	1	37	right,UIQ	3x2.5	
Ali et al. (1979)	2	57 42	left,UOQ left,UOQ	2 4	
Bogomoletz et al. (1981)	1	65	right,LIQ	3x2	
Gump et al. (1981)	17	18-80		1.5-10.0	
Pang & Alagaratnam (1982)	1	20	right,lower	3x1.5	
Cederlund et al. (1984)	1	28	left.UOQ	4x3x3	
Hanna et al. (1985)	1	39	left,UOQ	1x0.7x0.8	+
Present case	1	23	left,UOQ UIQ	4x4x1.5	+

*LIQ; lower inner quadrant, UIQ; upper inner quadrant
LOQ; lower outer quadrant, UOQ; upper outer quadrant

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