

Breast Hamartomas

An Important Entity in Mammary Pathology

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Summary. 1. Fifteen cases of a rather distinct entity of tumour-like lesion in the breast are presented.

2. They have a distinct mammographic appearance and can be easily shelled out at surgery, leaving symmetrical breasts.

3. They are well-circumscribed lumps of varying size with a smooth surface.

4. Microscopically they show normal or dysplastic glandular structures of lobules and ducts, without neoplastic features, within a prominent fibrous stroma. The lesions appear to be closely related to adenolipomas.

Key words: Breast — Hamartoma — Mastoma — Benign lesion.

Introduction

In the literature there are a few reports of well circumscribed “tumours” in the breast, characterized by normal or dysplastic breast tissue without neoplastic features. Prym (1928) in a comprehensive work, described such lesions as mastomas. The first conclusive clinical and histopathological description seems to be by Hogeman and Östberg (1963, 1968), who described three typical cases. There are two later reports (Arrigone et al., 1971; Hessler et al., 1978) in which the name hamartoma was proposed.

We report 15 cases, with details on clinical history, radiology, histopathology and therapy.

Material

Our material consists of 15 cases studied in the period 1962–1978. Most of these, 10, came from Malmö, a city of about 240,000 inhabitants. Five of the cases came from hospitals outside Malmö.

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Six of the cases were diagnosed the years 1976–1978 and three of them were found during a mammographic screening programme, involving women aged 50–70. Altogether 12,500 women have been screened. Cases 1–3 were described by Hogeman and Östberg 1968.

Data on age, site and history are to be found in Table 1.

Table 1

Case No.	Age	Site	Tumour size	Historical data and comments
1	40	left	17 × 9 cm (1,400 g)	Left breast larger than the right since puberty. Two normal deliveries 17 and 5 years earlier. After the last delivery the left breast grew larger and at admission was 4 times the size of the right. No palpable tumour but at operation a large tumour was easily enucleated leaving a normal-sized breast.
2	35	left	15 × 5 cm (840 g)	Two deliveries, the last seven years ago. After seven months lactation the left breast began to grow continuously and was about 3 times as large as the right at admission. A fist sized tumour was palpated and easily enucleated at operation.
3	20	right	7 × 5 cm	Noticed a swelling of the right breast during the last 3 months of gestation. The breast was markedly tender and lactation was impaired. The breast increased in size during the following 8 months. A well delimited tumour could easily be shelled out.
4	42	left	9 cm (140 g)	Left breast "always" larger. No children but an abortion mens II three years earlier. Last four months increase of the left breast. A soft tumour could be palpated behind the breast. Mammography: Circumscribed, dense tumour.
5	58	left	8 cm (132 g)	One child. No hormones. Admitted because of pain in her left breast. No tumour could be palpated, but mammography disclosed a well circumscribed tumour of high density, occupying the lateral half of the breast.
6	64	left	10 × 3 cm	Eleven years earlier operated in her left breast because of circumscribed tumour found by palpation at a screening examination. A local excision including part of the tumour was done. Histology: fibrosis. Now at screening mammography a large circumscribed tumour.
7	26	left	5 × 3 cm	One child 12 months old. After cessation of 6 months lactation the patient had a condition resembling mastitis in her left breast. Antibiotics without effect. Mammography revealed a rounded well circumscribed tumour of high density under the nipple.
8	52	right	3.5 × 1.5 cm	Two children. Eight years earlier conization because of cervical carcinoma-in-situ. Screening mammography revealed a rounded, well circumscribed tumour of high density beneath the nipple.

Case No.	Age	Site	Tumour size	Historical data and comments
9	30	left	16 cm (640 g)	Came for health control. The left breast had "always" been larger than the right one, but in the last year rather rapid growth had occurred. At palpation "grape-like" lumps were felt. At mammography circumscribed tumour, interpreted as cystosarcoma phyllodes. Fine needle aspiration: no malignancy. Mastectomy was performed on suspicion of cystosarcoma phyllodes. Centrally in the specimen a well circumscribed glistening tumour with small cysts.
10	15	right	13 × 9 × 4 cm	After menarche at 12–13 years she noticed a rapid growth of the right breast, that became double the volume of the left breast. At palpation a well circumscribed tumour, that could easily be enucleated.
11	45	left	8.5 × 7 × 3.5 cm	Growing left breast since unknown time. At operation a well circumscribed easily enucleated tumour between the mass of the breast and the pectoral fascia.
12	66	right	3.5 × 2 cm	Screening mammography revealed a carcinoma that proved to be a ductal infiltrating carcinoma. In the breast near the submammary groove was a well circumscribed tumour.
13	55	left	1 × 0.5 cm	Screening mammography revealed a well circumscribed tumour, thought to be a fibroadenoma. In a wedge excision there was a well circumscribed easily enucleated tumour.
14	39	left	5 × 4.5 × 1.5 cm	On the pill for a period 6 years ago. Two normal deliveries 4 and 1 year ago. Lactation history unknown. Now at mammography rounded, well demarcated tumour, that was excised by a wedge resection.
15	24	right	17 × 16 × 9 cm	Rapid growth of the right breast noted after two months on the pill. Suspicion of cystosarcoma phyllodes. Mammography: inconclusive because of the density of the tumour. Cytology: no malignancy. Because the patient was pregnant m.III only a biopsy of the well encapsulated tumour was done. Histology: cystic disease. Nine months later the large well demarcated tumour was excised.

Results

Clinical Picture. All patients were females, age range 15–66 years. Ten of the 15 tumours were left-sided.

Five of the cases were noted in connection with lactation. There was a history of long duration in most cases indicating a very slow growth. Some of the patients (cases 1, 2, 3, 9 and 10) presented with very large "tumours" giving a striking asymmetry of the breasts (Fig. 1), but four cases with rather large "tumours" (up to 8 cm) were incidental findings at mammography. Six

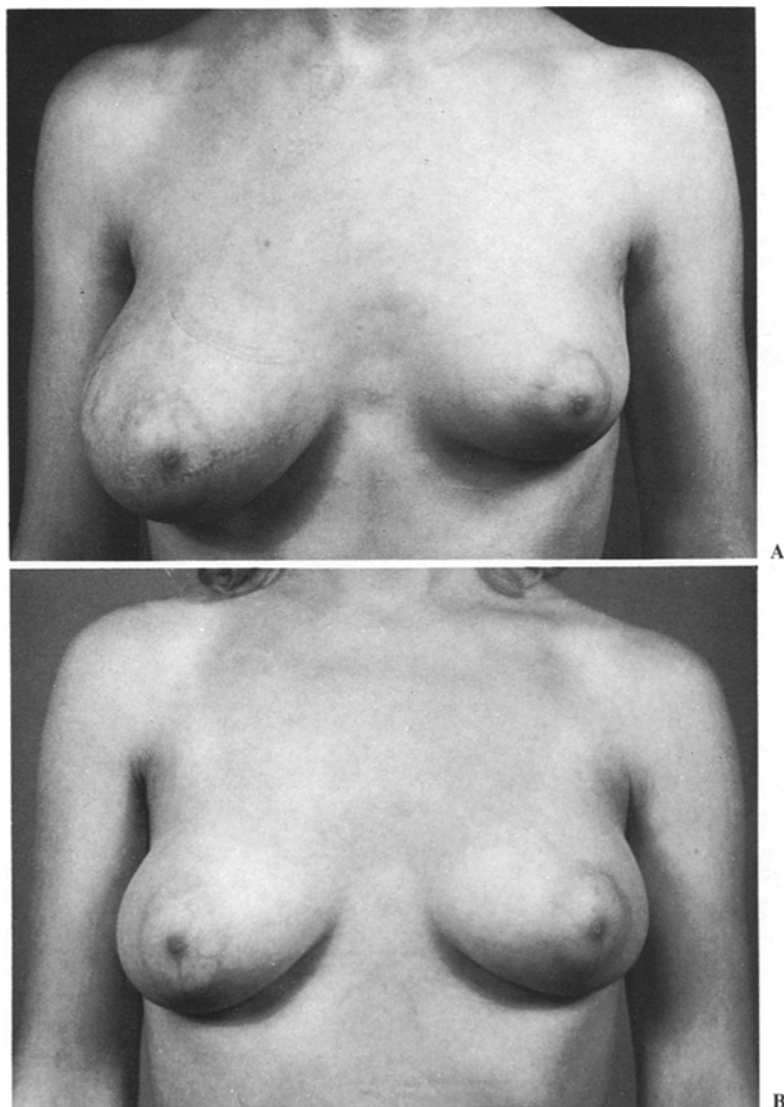


Fig. 1. Case 10. A. Before operation. B. After operation with symmetrical breast

of them could not be palpated, probably because they were often located behind the normal breast parenchyma.

Most cases had no subjective symptoms but when the "tumours" were very large they were occasionally painful and gave a sense of heaviness. One case (Nr. 7) was regarded as mastitis.

Diagnosis. Some of the lumps could be palpated, especially the large ones. Mammography usually gave a very distinctive picture of a well circumscribed,

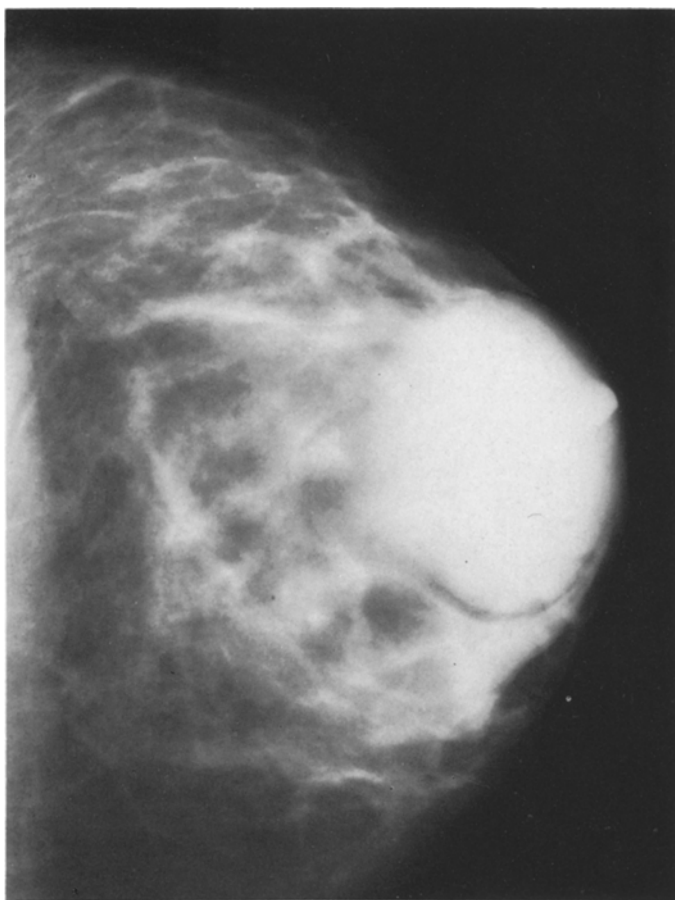


Fig. 2. Case 7. Mammographic picture of a 3 cm big lens-shaped very dense and well delimited tumour beneath the mamilla. The radiolucent zone is readily seen especially to the lower side

rather dense, mostly round or lens-shaped mass surrounded by a narrow radiolucent zone (Fig. 2). Small lesions could not be differentiated from cysts or fibroadenomas.

Surgical Approach. The lumps were usually easily shelled out from a submammary incision. They were surrounded by very loose tissue and could easily be freed by blunt dissection (Fig. 3). There was practically no bleeding. In one case (Nr. 10) there was a slender stalk of normal mammary tissue connecting the lump with the rest of the breast.

After enucleation of the lump the mass of mammary tissue remaining roughly equalled the amount on the opposite side (Fig. 1). In one case (Nr. 9) a mastectomy was performed on a false suspicion of cysto-sarcoma phyllodes.

Pathological Anatomy. The lesions varied greatly in size. The largest (case 1)



Fig. 3. Case 4. A 9 cm large tumour with smooth surface. The cut surface shows oedematous glistening parts

weighed 1,400 g with a diameter of 17 cm. The smallest (case 14) had a diameter of 1 cm. All had a smooth and even surface (Fig. 3) with some adhering remnants of the surrounding loose connective tissue. The consistency was firm but not hard. The cut surface was mostly grey and glistening (Fig. 3) and some small cysts could be seen (Fig. 8). The tissue of the lumps was occasionally rather oedematous (Figs. 3, 4). A remarkable feature was the absence of the bulging lobular elements so characteristic of fibroadenomas. Phylloid splitting, as seen in cystosarcoma phyllodes was not present.

The histological appearance was characterized firstly, by the absence of adhering adjacent breast tissue, as often seen in fibroadenomas. These tumours had no real capsule and the surface looked naked (Figs. 4–7). They consisted mainly of fibrous tissue, often rich in capillary vessels, with scattered glandular elements of ordinary appearance with both lobules and ducts (Figs. 4–7). In a few cases (Fig. 6) numerous glandular elements were seen but in most instances the fibrous stroma predominated (Figs 5, 7). In some cases dysplastic changes were seen in cystic ducts with apocrine epithelium that was sometimes papillary. Severe epitheliosis was never seen. The distorted lobular structures characteristic of fibroadenomas were never observed. Areas of fatty tissue were sometimes seen but the amount was small in most of our cases (Fig. 7).

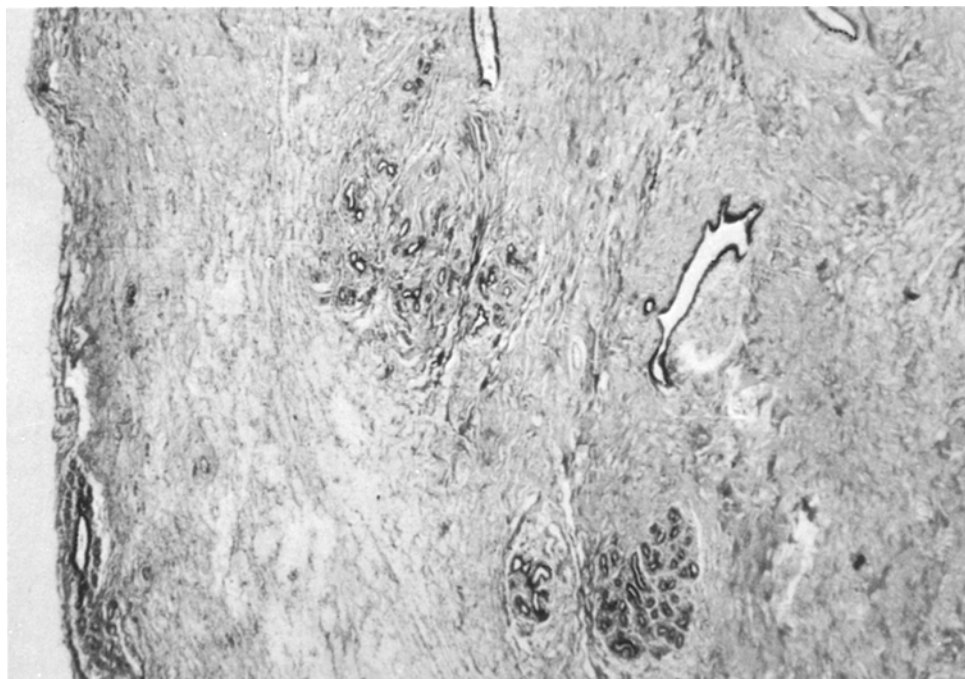


Fig. 4. Case 10. 15 year old with 13 cm large tumour. To the left the smooth surface. The stroma is dense fibrous with oedematous loosening to the left. A few unevenly distributed lobules and ducts. $\times 50$

Discussion

The lesions described here constitute a well defined entity with the following main features:

1. Well circumscribed lumps of variable size from 1 cm to 17 cm in diameter (the largest weighing 1,400 g).
2. At physical examination some tumours cannot be palpated, while others are felt as lens-shaped masses, separated from the rest of the parenchyma.
3. At mammography they appear as well circumscribed, round or lens-shaped rather dense lesions, mostly surrounded by a narrow radiolucent zone.
4. At operation they are very easily enucleated without bleeding; after enucleation the breasts are symmetrical.
5. The lumps consist of normal or occasionally dysplastic mammary tissue without neoplastic properties. Fibrous stroma predominates. To a greater or lesser extent fatty tissue can also be found. A well delimited macroscopical appearance is the most typical feature.

The fifteen cases constituting this report were found during a period of 15 years. However, 8 of our cases were diagnosed in the last two years, most of them representing incidental findings at mammography. This type of lesion seems therefore to be much more common than anticipated. The literature

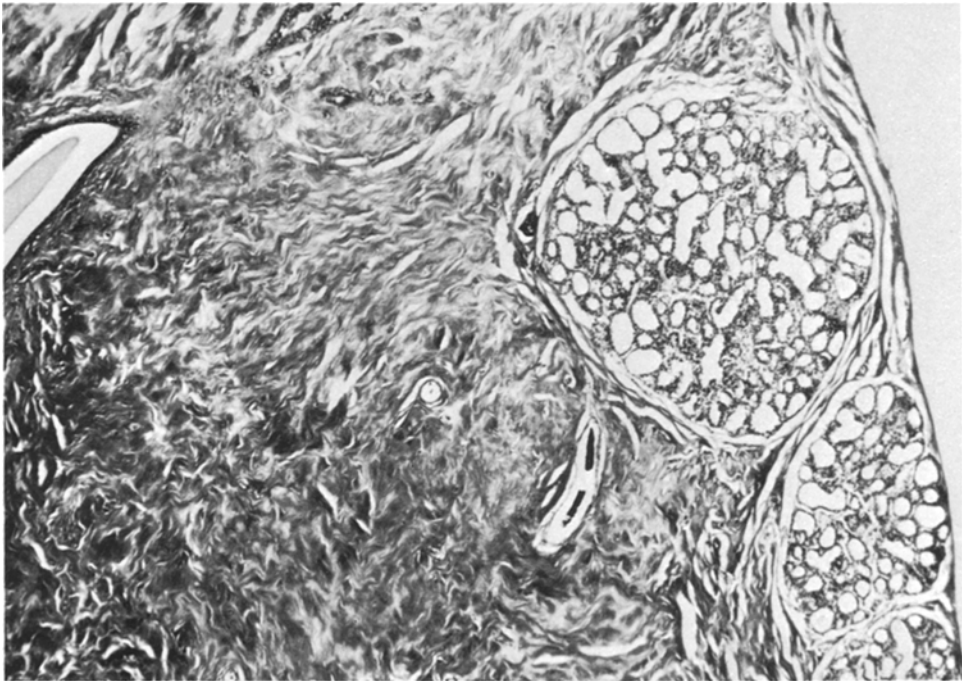


Fig. 5. Case 14. Extremely dense fibrous stroma. To the right just beneath the smooth surface a few lobules with secretory changes. $\times 50$

contains few reports. Prym (1928) in a very comprehensive study on fibroadenomas and "mastomas" described similar cases. He called them mastomas and stressed that they were built up of normal or dysplastic mammary tissue and had, in contrast to fibroadenomas, no connection with the surrounding breast parenchyma. He also pointed out that they sometimes were noticed after lactation. He gave the very plausible interpretation that these masses of separated breast tissue, without connection with the ducts, showed slower post-lactational involution. Hogeman and Östberg (1963, 1968) described three cases and gave the first clear description of the macro- and microscopical picture. They described the lesions as a peculiar type of postlactational breast tumour, because all their cases were noticed after lactation. Arrigoni et al. (1971) described ten cases and illustrated the typical anatomical structure. They accepted that pregnancy and lactation could influence the rate of growth. Hessler et al. (1978) described 16 cases found by mammography but denied any connection with lactation. We think that these lesions are not caused by lactation, but are modified by it occasionally growing faster after lactation. So called pure or lactating adenomas probably belong to the same group of lesions, consisting of a well delimited part of lactating breast parenchyma.

Arrigoni et al. proposed the name hamartoma. Although we think that these lesions do not exactly correspond with the definition of hamartoma as given by Albrecht (1906), we are of the opinion that the name hamartoma now

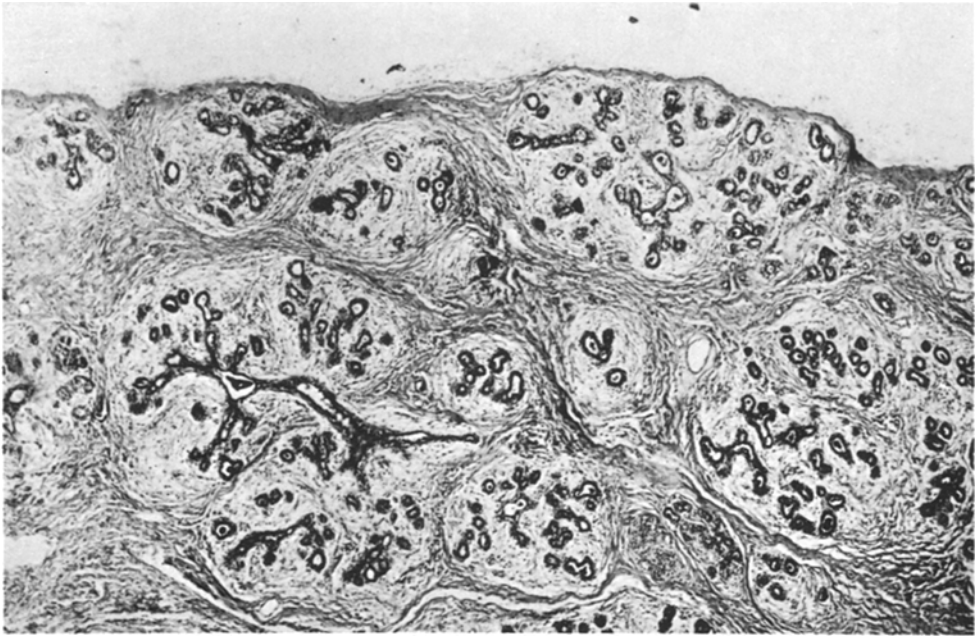


Fig. 6. Case 7. Rather many lobules with loose stroma. In the upper part the smooth surface. $\times 50$

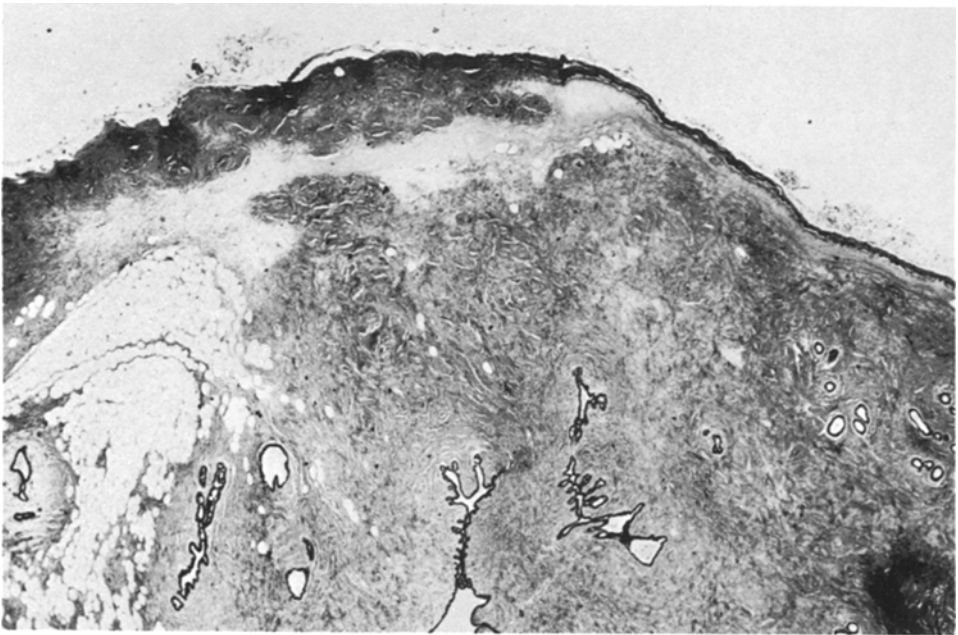


Fig. 7. Case 11. In the upper part oedematous stroma beneath the smooth surface. To the left fatty tissue. $\times 22$



Fig. 8. Case 16. Well circumscribed tumour. The cut surface shows several cysts

has been widely adopted and therefore its use is justified. Prym's proposal to call them mastoma may be a good alternative.

Clinical diagnosis is not often possible. However, the finding at mammography of a lens-shaped mass, separated from the rest of the parenchyma, is suggestive of the diagnosis. Only when the surgeon enucleates the whole tumour does the pathologist get the opportunity to see the typical macro- and microscopical picture. We imagine that in the past many such cases have been partially resected and that the pathologist receiving such a specimen has given the diagnosis of fibrosis or fibroadenosis. Our case 6 is one such example. Small pieces of these "tumours" appear wholly uninteresting and banal to the pathologist. Without doubt, mammography has contributed to the increased numbers found during the last two years.

Mammary hamartomas are seen in all ages after puberty. Our youngest case was 15 years and had a very fibrous tumour (case Nr. 10) (Figs. 1, 4). This seems to indicate that these masses of breast become separated during development, perhaps in fetal life. It would be interesting to look for these lesions in cases of gynecomastia.



Fig. 9. Case 16. To the left the smooth surface. Cellular stroma with some hyperplastic lobules. $\times 50$

Spalding (1945) first described adenolipoma and Haagensen mentioned 22 own cases. Adenolipomas are well circumscribed tumours consisting of fatty tissue containing variably glandular tissue lying “naked” in fat. Spalding regarded adenolipomas as tumours “*sui generis*”. The hamartomas, described by us and others, occasionally contain small or large parts of fatty tissue [see, for example, case 11 (Fig. 7)]. There is good reason to believe that there is a continuous variation from fibrous and glandular hamartomas to typical adenolipomas. Hessler et al. (1978) also grouped them together. Menges et al. (1976) gave a good description of one case.

The aetiology is obscure. Probably these hamartomas are to be looked upon as developmental anomalies that can be influenced by hormones, for instance at lactation. Our patients did not use hormones but two were on the pill for a short while.

The anatomical picture points to a non-neoplastic nature. No recurrences are described and the lesions seem to be wholly benign. However, we have seen a peculiar case that resembles the hamartoma but has recurred twice. This was a 28 year old woman, who had palpated a lump in the left breast. At surgery a well circumscribed partly cystic tumour was enucleated, the size of “half a mandarine” (Fig. 8). Six years later a new lump was palpated under the scar and a well circumscribed nodule of 28×22 mm was enucleated. Sixteen months later a third well circumscribed “golf ball sized” lump was excised. Observation time after last surgery is only one month.

The anatomical picture was the same in all three instances. Macro- and microscopically the tumours were sharply delimited (Fig. 8). The stroma (Fig. 9) was very cellular but without atypia or mitoses, resembling that seen in some giant fibroadenomas (cystosarcoma phyllodes), but without phylloid splitting. The stroma contained normal lobules and ducts, with focally hyperplastic but regular epithelium.

This case has much in common with the other cases, but the recurrences are remarkable. At each instance the surgeon had the impression of having enucleated a well demarcated tumour, leaving no remnants behind. We do not know if this case is a variant of the other cases or represents a special type. The possibility of multiple "Anlage" may also be considered. A follow-up of the patient will show the real biological behaviour of this tumour.

The proper therapy for large or otherwise uncomfortable hamartomas is enucleation, which gives a good cosmetic result with almost symmetrical breasts (Fig. 1). Hessler et al. stress that the mammographic appearance is so distinctive that in selected patients presenting a clear picture of hamartoma, the lesion may be left when it is small and does not cause discomfort.

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