CASE REPORT

Ductal carcinoma in situ arising in mammary hamartoma

G M K Tse, B K B Law, L M Pang, H S Cheung

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Hamartoma of the breast is an uncommon lesion. Although it can possess characteristic radiological features, the pathological appearance is not distinctive. Hamartoma is generally considered benign, but four cases have been reported with ductal and lobular carcinoma arising in hamartomas. This report describes further cases of hamartoma from which ductal carcinoma in situ arose, with one showing early invasion. In both cases, the tumours were within the hamartomas and were adequately excised during lumpectomies of the hamartomas, and the patients were well afterwards. This report emphasises the importance of adequate sampling of mammary hamartoma.

CASE REPORTS

Case 1

A previously healthy 32 year old woman presented with a three month history of a right breast lump. Clinical examination revealed a 1.5 cm mobile firm lump at the right upper outer quadrant with no contralateral or axillary abnormality. The clinical diagnosis was fibroadenoma. Ultrasound examination showed a 2 × 0.9 cm well defined lobulated mass at right 10 o'clock position. Mammography was not performed. Fine needle aspiration cytology of the lump yielded a benign aspirate. The mass was excised and the patient remains well and asymptomatic with five year follow up.

Case 2

A 68 year old postmenopausal woman presented with a two month history of a right breast lump. Physical examination revealed a 1.5 cm lump at the right upper outer quadrant with no contralateral or axillary abnormality. Mammography was not performed. Fine needle aspiration cytology of the lump revealed cellular atypia. The lump was excised and she remains well and asymptomatic with four year follow up.

Figure 1 Low power photomicrograph of case 2 showing a hamartoma with rounded border and a fibrous capsule. Haematoxylin and eosin stained.

PATHOLOGY

In case 1, the specimen measured 1.5 cm in largest diameter. It possessed a smooth surface and was well circumscribed. The cut surface showed focal islands of yellow fatty tissue. Microscopically, the background was fibrotic with focally dense fibrous tissue encasing the adipocytes. Only very rare ducts were present. A 0.7 cm focus of ductal carcinoma in situ (DCIS) was seen, with the ducts being distended by a monotonous epithelial cell population. The cells were arranged around the circumference of the ducts, focally forming infoldings to give a micropapillary pattern. There was a mild degree of nuclear pleomorphism, and an intact layer of myoepithelial cells could be discerned. The DCIS was within the hamartoma and did not extend to the margin of the resection.

Figure 2 A focus of ductal carcinoma in situ in case 2 with solid and cribriform pattern. Normal lobules are seen at the edge of the picture (asterisks). Haematoxylin and eosin stained.

Figure 3 A focus of ductal carcinoma in situ of solid type in case 2. Isolated tumour clusters are seen free in the stroma within a mucinous background. Microcalcification is also noted (arrow). Haematoxylin and eosin stained.

In case 2, the specimen was a yellow to tan nodule, measuring 2.5 cm in largest diameter, with a rounded border and fibrous capsule (fig 1). The cut surface was uniform in colour. Microscopically, many ducts and lobules were present within a fibrotic stroma, and some of these were cystically dilated with apocrine metaplasia. No compression of the ducts or lobules to slit-like spaces was seen. Some adipose tissue and oedema were noted in the background. A 0.8 cm focus of DCIS was seen (figs 2,3). There was distension of the ducts by a uniform population of malignant cells with mildly pleomorphic nuclei and occasional mitoses. Within the DCIS, a 0.1 cm invasive focus was noted, with nests and tubules of similar tumour cells floating within a collection of extracellular mucin, consistent with mucinous carcinoma. The tumour was within the hamartoma and well away from the resection margin.

DISCUSSION

Hamartoma of the breast was first described and the term coined in 1971 by Arrigoni.¹ In the literature, several large series have reported more than 120 cases,²⁻⁶ all of which were considered benign. Four morphological patterns have been described, including encapsulated breast parenchyma with fibrocystic changes, fibroadenoma like with fibroblastic stroma surrounding glandular structures, fibroadenoma like with normal lobules, and circumscribed adenolipoma with mostly adipose tissue containing scattered ducts and lobules.² However, these morphological divisions have not been widely used. The mammographic appearance of some hamartomas is characteristic, seen as a well circumscribed encapsulated mass density with lobulated or smooth borders containing fat density.⁷ However, because a distinct pathological picture is absent, hamartoma may be under-recognised by pathologists without the benefit of imaging, particularly in fine needle aspiration specimens or in needle core biopsies.⁵ The major differential diagnosis is fibroadenoma, although hamartomas possess more glands and lobules, more fat, and less stromal cellularity than fibroadenoma.

"Until the association between malignancy and hamartoma is better established, the practising pathologist should examine the specimen diligently and sample adequately"

Malignancy arising from or within a hamartoma is exceedingly rare, only four cases have been reported in the English literature,⁸⁻¹⁰ including a report of two cases of infiltrating duct carcinoma, one of which was found entirely within a hamartoma.⁸ Another paper reported lobular carcinoma in situ with focal invasion within a hamartoma.⁹ A recent report described a case with high grade and extensive infiltrating and in situ duct carcinoma with lymph node metastases.¹⁰ The authors report two additional cases of low grade DCIS, with one containing a focus of early invasion present entirely within the hamartoma. With the addition of these two cases to the literature, the whole spectrum of ductal and lobular carcinoma, both invasive and in situ, has now been reported. A parallel situation is seen in fibroadenoma. The incidence of malignancy arising in fibroadenoma is reported to be less than 0.5%, with lobular carcinoma in situ occurring in about half of these cases, and invasive ductal, lobular, and in situ ductal carcinoma occurring in roughly equal proportions for the remaining half. In 50% of these cases, the malignancy extends beyond the fibroadenoma. The reported age for malignancy in fibroadenoma is higher than that for benign fibroadenoma. For hamartoma, which is still under-recognised, the occurrence of malignancy is rare. A review of the larger series in the

Take home messages

- Hamartoma of the breast is an uncommon lesion that is probably underdiagnosed—with the increasing use of mammographic or clinical screening, more hamartomas will most likely be identified and diagnosed
- Although malignancy arising from or within a hamartoma is exceedingly rare, it can occur
- Thus, adequate sampling of mammary hamartoma is essential

literature of benign hamartomas where patients' ages were reported^{2-4, 6} gave a mean age of 37.8 (range, 13 to 89); for the cases with malignancy,⁸⁻¹⁰ including those in this report, the mean patient age was 58.2 (range, 32 to 78). The difference is significant (student's *t* test, *p* = 0.001). It is expected that with the increasing use of mammographic or clinical screening, more hamartomas will be identified and diagnosed. Increased awareness of the possibility of malignancy arising in hamartoma would lead to a higher incidence. No long term follow up data on this group of patients are available in the literature, but in the case reports, patients with infiltrating carcinomas in hamartomas had mastectomy and axillary dissection,^{8, 10} and the patient with predominately in situ carcinoma had local excision.⁹ It appears that treatment is directed towards the coexisting malignancy, and not the hamartoma itself. Until the association between malignancy and hamartoma is better established, the practising pathologist should examine the specimen diligently and sample adequately.

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Authors' affiliations

G M K Tse, Department of Anatomical and Cellular Pathology, Prince of Wales Hospital, The Chinese University of Hong Kong, Hong Kong SAR
B K B Law, Department of Surgery, Prince of Wales Hospital
L M Pang, H S Cheung, Department of Diagnostic Radiology and Organ Imaging, Prince of Wales Hospital

Correspondence to: Dr G M K Tse, Department of Anatomical and Cellular Pathology, Prince of Wales Hospital, Ngan Shing Street, Shatin, NT, Hong Kong; garytse@cuhk.edu.hk

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