Low and high grade mucoepidermoid carcinomas of the breast

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Summary

Mucoepidermoid carcinoma is a very rare primary tumour of the breast. Until now only 17 cases have been described in the literature. Generally these malignancies have a good prognosis, especially if well differentiated. We report a case of low-grade which recurred as high-grade after 32 months. She also developed a poorly differentiated adenocarcinoma in the other breast 12 years after her initial treatment. At present the patient is well without any sign of disease for 156 months. A literature review of this rare entity is presented.

Key words: Mucoepidermoid carcinoma; Breast cancer; Pathology; Prognosis.

Introduction

Mucoepidermoid carcinoma comprises between 2.8% and 15.5% of all salivary gland tumours, between 12% and 29% of malignant salivary gland tumours and between 6.5% and 41% of minor salivary gland tumours, representing the most common type of malignant minor salivary gland tumour [1]. Primary mucoepidermoid carcinoma of the breast however, is extremely rare with an estimated incidence of 0.2-0.3% [2]. In the literature we did not find any information about the radiological appearance of this tumour. It was first described by Pachevsky *et al.* in 1979 [3] and to date only 17 cases have been reported [2-12].

A case of low-grade mucoepidermoid which recurred as high-grade while the patient also developed a poorly differentiated adenocarcinoma in the other breast 12 years after her initial treatment is presented, together with a review of the literature.

Case Report

At age 58 the patient presented with a mobile mass in the right breast. On mammography an inhomogeneous mass with obscured margins deep in the upper, outer part of the right breast was seen. Sonography revealed an inhomogeneous mass, 3.5 x 3 cm, with unsharp margins. The patient was diagnosed with a low-grade mucoepidermoid carcinoma (Figure 1).

Mastectomy and axillary clearance were performed according to Patey. One of the 17 removed lymph nodes was involved by metastatic tissue. The oestrogen and progesterone receptors were both negative. The tumour was staged as pT2pN1MO and the patient received adjuvant radiotherapy (50 Gy) and hormonal therapy (tamoxifen 20 mg daily). After a disease-free period of 16 months a local recurrence (3 nodules with diameters of 1.5, 0.7, and 0.4 cm) occurred with infiltration in the major pectoralis muscle. Treatment consisted of wide local excision and adjuvant radiotherapy of 40 Gy. The latter due to the fact that the dorsal margin was positive for tumour cells. After 32 months a re-recurrence occurred (4 x 3 cm), without evidence of distant metastasis. This time the tumour appeared to be high-grade instead of the previously low-grade (Figure 2). A resection of the thorax wall was performed as previously described by Tjalma *et al.* [13]. Shortly after, resection of rib IV until VI (area 17 x 13 cm), closure of the thoracic wall defect with a polyethylene mesh and a soft tissue reconstruction with a myocutaneous lattissimus dorsi flap were done. No further treatment was given. At age 70 the patient developed a poorly differentiated breast cancer in the remaining breast. Mastectomy with axillary clearance was performed and the tumour was staged as pT1pN0M0. The oestrogen and progesterone receptors were both negative. No further adjuvant therapy was given. At present, 156 months after the initial treatment, the patient is well with no sign of disease.

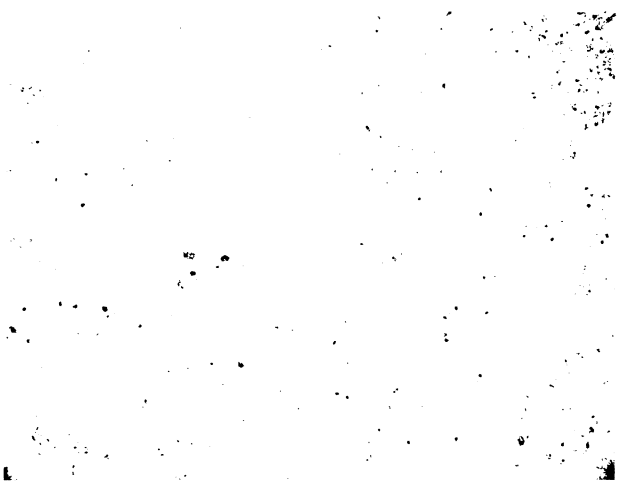

Figure 1. — PAS-stain of low grade mucoepidermoid carcinoma of the breast, showing cohesive epithelial strands with a secretory vacuole containing neutral mucins in some cells. Cellularity is moderate and cellular pleiomorphism not very marked.

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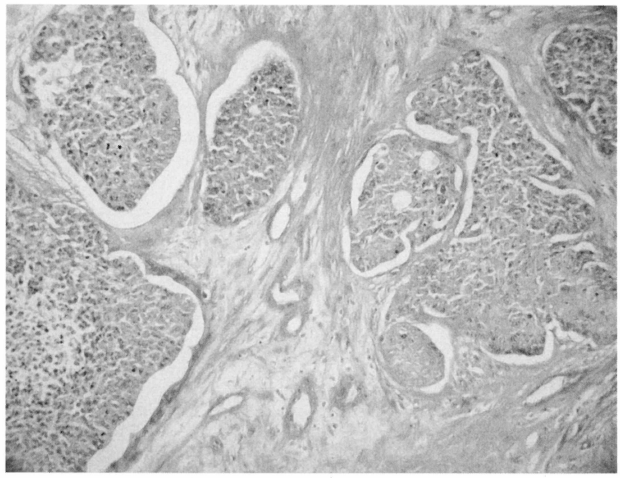

Figure 2. — PAS-stain of high grade mucoepidermoid carcinoma. In contrast to the tumour in Figure 1, cellularity is high with marked hyperchromasia of the nuclei. Large areas of necrosis are also conspicuous.

Discussion

Mucoepidermoid carcinomas are malignancies that occur mainly in the submandibular and parotid glands. However, they have also been reported in the bronchus, oesophagus, lacrimal glands, conjunctiva and penis [11].

Microscopically mucoepidermoid carcinomas are composed of mucus cells containing neutral mucins, intermediate cells with a basaloid appearance, squamous cells and clear cells. The differential diagnosis for mucoepidermoid carcinomas includes narcotising sialometaplasia (NSM), metastatic squamous carcinoma (for high-grade tumours), adenosquamous carcinoma, cystadenoma, cystadenocarcinoma, sebaceous carcinoma, and other clear cell carcinomas, oncocytoma, oncocytic carcinoma, and metastatic renal cell carcinoma [1].

These tumours are histologically classified into low, intermediate and high grade based on the relative proportion of cell types. A high grade tumour is solid and contains predominantly intermediate and squamous cells, while a low grade tumour is composed of a higher proportion of mucus cells [1, 11]. Not surprisingly the mucoepidermoid carcinomas are the most frequently misdiagnosed lesions by fine needle aspiration (FNA), leading to a delay in diagnosis and treatment [11].

The prognosis of mucoepidermoid carcinoma in the salivary glands is indicated by grade, completeness of excision and clinical staging. The recommendations in the salivary glands are no additional lymph node dissection in grade 1 tumours; in grade 3, lymph node dissection is included as standard therapy, while grade 2 tumours are left to the surgeon's discretion. Low-grade tumours have a 5-year survival rate of 90-100%, while high grade lesions have a survival rate of almost 50%. One could assume that this also holds true for breast cancers. A review of the literature (Table 1) confirms the suspected correlation of grade, lymph node metastases and survival [2-12].

The present report is unique in the sense that the low grade tumour recurred twice and the second time as a high grade lesion. In addition, this is the first low grade tumour with a metastatic involved lymph node. The patient developed a second primary tumour in the other breast. The current case sheds light on tumour histiogenesis. Theoretically this tumour can have a multicellular origin or arise from a single clone of pluripotent cells. The single cell clone theory is based on the assumption that the intermediate cells represent transitional cell

Table 1. — *Reported cases of primary mucoepidermoid carcinoma of the breast.*

	Author	Year	Age	Grade	LN	Status	Follow-up
1.	Pachevsky *et al.*	1979	66	Low	0/20	NED	8 years
2.	»		70	Low	--	NED	10 months
3.	Kovi *et al.*	1981	46	High	17/19	--	--
4.	Fisher *et al.*	1983	65	Low	--	NED	5 years
5.	»		71	Low	0/19	NED	4 years
6.	»		57	Low	0/11	NED	10 years
7.	»		49	Low	0/13	NED	9 years
8.	»		60	Low	--	DOID	4 years
9.	Ratanarapee *et al.*	1983	27	High	6/15	DOD	14 months
10.	Leong *et al.*	1985	57	High	0/20	DOD	7 months
11.	Hanna *et al.*	1985	51	--	0/--	NED	8 months
12.	»		31	--	2/18	NED	18 months
13.	Hastrup *et al.*	1985	59	High	0/4	DOD	25 months
14.	Luchtrath *et al.*	1989	60	High	12/18	DOD	30 months
15.	Pettinato *et al.*	1989	72	High	16/19	DOD	10 months
16.	Markopoulos *et al.*	1998	40	High	0/--	NED	5 years
17.	Berry *et al.*	1998	51	High	0/--	NED	--
18.	Tjalma *et al.*	Present	58	Low*	1/17	NED	13 years

NED = no evidence of disease; DOID = death of intercurrent disease; DOD = death of disease; -- = not mentioned; * = recurrence as high grade.

forms between duct cells and myoepithelial cells or between squamous cells and myoepithelial cells. While the multicellular origin of these tumours is based on the consideration that they have ductal and myoepithelial derivation [7], despite this pattern of tumour development the patient is well and alive 13 years after her initial treatment.

In conclusion mucoepidermoid carcinomas of the breast are very rare. The treatment should be as for other breast malignancies. The overall prognosis for these breast tumour types is good.

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