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CLINICAL CASE

Adenomyoepithelioma of the breast; a case report and literature review

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Summary

A case of adenomyoepithelioma of the breast is presented in order to illustrate some of the difficulties in achieving a pathological diagnosis of this lesion. Given the emerging evidence for adenomyoepithelioma to develop into malignancy, it is imperative that the histopathological features of this lesion are well described and recognized.

Key words: adenomyoepithelioma, breast, histopathology

Introduction

Adenomyoepithelioma of the breast is a rare tumour first described in 1970[1]. Similar tumours derived from myoepithelial cells are well recognized in the salivary gland. They are typically characterized by biphasic proliferation of both epithelial and myoepithelial cells. However, the specific histological features of adenomyoepithelioma are variable and subtle and can subsequently lead to erroneous diagnoses.

Although most breast adenomyoepitheliomas are benign [2,3], potential for malignant evolution of this lesion has been described [2,4,5] with subsequent presentation either as a local recurrence or, rarely, with distant metastases [6-8]. It is therefore important that the correct diagnosis is reached to ensure optimal clinical management of the patient.

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Case presentation

A 56-year-old woman presented with a three-week history of a discrete lump and pain in the left breast. She had been taking hormone replacement therapy following a bilateral salpingo-oophorectomy in 1992. Two aunts and a female cousin had a history of breast cancer.

Clinical examination revealed a 1 × 1.5 cm poorly-demarcated, non-painful lump in the lower medial quadrant of the left breast. There was no fixity of the lesion to underlying skin or to muscle and no lymph nodes were palpable. Mammography was normal in both breasts but ultrasound of the left breast revealed an ill-defined solid mass 18 mm in diameter that was homogenous and hypoechoic in nature.

Core biopsy showed an unusual tumour composed of diffusely infiltrating strands and islands of cells formed from a biphasic population of epithelial and myoepithelial cells (Figure 1). Around the tumour, and within pseudoacini, there was abundant basement membrane material. The appearances were suggestive of a salivary gland type tumour.

An expert breast histopathological opinion was sought (IOE). They confirmed that the specimen showed a variable morphological appearance; for the most part there was an adenotic, organoid structure suggesting that the lesion involved expanded terminal duct lobular units. In other areas there was a nodular configuration with low power appearance of an underlying papillary intraductal proliferation. Both ar-

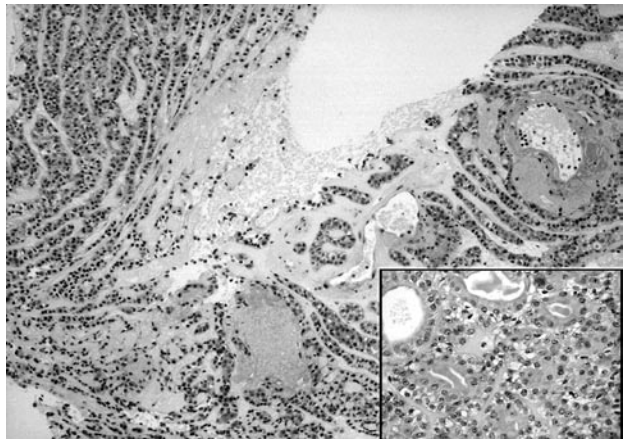

Figure 1. Core biopsy showing cords of cells with scanty tubular structures (H&E x100). At higher power (inset) the cells are of moderate size with prominent nucleoli (H&E ×200).

eas contained a mixed cell population, although there was a dominant small, relatively uniform cell type. These latter cells had irregular nuclei with prominent nucleoli and a moderate amount of clear cytoplasm, although some had more eosinophilic cytoplasm.

Immunophenotyping confirmed that there was a dual population of myoepithelial (smooth muscle actin and smooth muscle myosin-positive, Figure 2) and epithelial (Cam5.2 positive) cells. In addition, there was a distinct surrounding basement membrane component (Figure 2 inset). Some of the epithelial islands had luminal spaces which also contained basement membrane material. Thus histological differential diagnosis rested between an adenoid cystic carcinoma and an adenomyoepithelioma. In view of the heterogeneity of architectural pattern, presence of an intraductal papillary component and the mixed phenotype of myoepithelial and luminal epithelial cells, the favoured diagnosis on core biopsy was an adenomyoepithelioma.

The patient underwent wide local excision and sentinel lymph node biopsy. Final histology confirmed the composition of predominantly myoepithelial cells with a small epithelial component. The tumour cells showed no significant pleomorphism. The lesion was also noted to arise from a benign intraductal papilloma measuring 11 mm in maximum diameter. No features of malignancy or vascular invasion were identified. The sentinel lymph node showed no evidence of tumour on both conventional haematoxylin & eosin and immunohistochemical stains. The features described were those of an adenomyoepithelioma.

She entered a 5-year follow up programme and, when last seen at 18-month follow up, there was no evidence of recurrence.

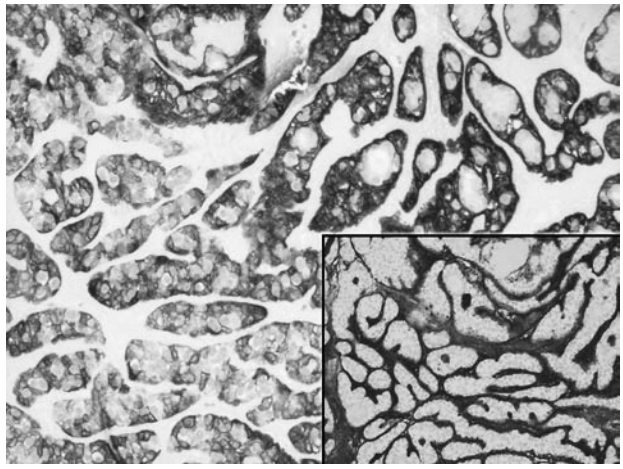

Figure 2. Main image: smooth muscle myosin immunohistochemistry shows the myoepithelial phenotype of many of the cells in the trabecular structures with the tubular structures and scattered cells negative (×200). Inset shows laminin positivity of the basement membrane material (×100).

Discussion

Adenomyoepithelioma is a rare lesion, which is classically benign, both histopathologically [2] and in radiological appearance [9]. Up to 35 cases have been recorded, in which the tumours were considered to be potentially malignant [8]. Conversely, it is important to note that cases of benign adenomyoepithelioma of the breast may mimic malignancy [10]. Care must therefore be taken in the diagnosis of adenomyoepithelioma, particularly if there is cytological atypia, and the lesion should not be misdiagnosed as infiltrating ductal/no special type carcinoma. Reaching the correct histopathological diagnosis is a potential problem with the limited sampling inherent in the use of core biopsy for preoperative diagnosis [11].

Typically, adenomyoepitheliomas are predominantly solid in architecture, although in a minority of lesions tubular structures are prominent. The diagnostic feature of the lesion is the identification of two cells types; a basaloid or spindled cell population, sometimes with clear cytoplasm which shows a myoepithelial immunophenotype (e.g. smooth muscle actin or smooth muscle myosin positivity) is seen along with a second population of cells with cuboidal morphology and which immunophenotypically are of glandular type and which form the inner tubular structures. Most commonly, the basaloid spindled cell

component predominates and the lesion has a solid appearance. A classification system proposed by Tavassoli, subdivides adenomyoepithelioma into 4 types: spindle cell, tubular, lobular and carcinoma arising in adenomyoepithelioma [12]. It is suggested that the different histological types predict for different presentation and clinical behaviour. However, it should be noted that the clinical behaviour of these lesions is difficult to predict by histological morphology.

Malignant change may involve only one cellular component, reportedly more often the epithelial than the myoepithelial portion [2,4], but the morphological features that predict the potential for local recurrence and/or metastasis are not well established [12]. No well-defined histological criteria can be used to define an individual lesion as benign or malignant. Malignant tumours have been described in the literature as exhibiting features such as a spindle cell component, local infiltration, an increased mitotic rate of more than 5 per 10 high power fields, or cellular atypia [3,5,12]. It has also been suggested that, in general, a diagnosis of malignancy is not warranted if nuclear atypia is not severe. However, given the potential for malignant clinical behaviour, it is of paramount importance that the correct histopathological diagnosis is made both for prognosis and treatment. The potential for local and distal recurrence has implications for surgical treatment and follow up; these lesions should be widely completely excised.

The above case illustrates the complexities of making the diagnosis of adenomyoepithelioma and care should be taken in the histological assessment of such lesions, particularly when suspected in the limited tissue sampling which is inherent in core biopsy interpretation.

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