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Intracystic Malignant Adenomyoepithelioma of the Breast: A Case Report and Review of Literature

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1. Abstract

Adenomyoepithelioma (AME) of the breast is a rare neoplasm with a biphasic proliferation of epithelial and myoepithelial cells. The authors report an unusual case of AME of 51-year-old woman. The AME was located in a cystic structure 6 cm in diameter as a solid nodular lesion with 2cm size. Immunohistochemically, the nodule was comprised with 2 cell population of tubular structures. First population of the glandular epithelial cells with dark cytoplasm showed the activity of CK7(+), CAM5.2(+), CK5/6(partly +), p63(-), CD10(-), S-100(-), αSMA(-). Second population of outer myoepithelial cells with clear cytoplasm displayed the activity of p63(+), CD10(+), CK14(+) CK5/6 (partly +), CK7(weakly +), CAM5.2(weakly +), S-100(+), αSMA(+). Beside them, cellular cluster of the inner epithelial cells with prominent nuclear as well as cellular atypia, and atypical spindle cell proliferation emanating from the myoepithelial component were also present. Thus, the nodule was diagnosed as malignant AME. One year after the lumpectomy, pulmonal metastasis of the breast tumor was also found. So far as we are concerned, such case of intracystic malignant AME of the breast has never been described. Regarding histogenesis of the neoplasm growing under the unusual environment, a consideration with review of literature was done.

2. Introduction

Adenomyoepithelioma (AME) was first described by Hamperl [1] in 1970 as a neoplasm consisting of both luminal and myoepithelial cells. Tavssoli subdivided AME as myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma [2]. Following statement of the current World Health Organization (WHO), AME is simply divided into two types of AME and AME with carcinoma [3]. Because of the biphasic nature of the tumor, malignant transformation may arise from the glandular epithelium, myoepithelium, or both [4-6] and the proportion of myoepithelium and glandular epithelium proliferation differs from case to case [6]. Thus, immunohistochemical features of AME and AME with carcinoma highlight their dual epithelium and myoepithelial composition [7]. As a myoepithelium component of AME, spindle cell proliferation is recognized [3].

Furthermore, in cases of AME with distant metastases, myoepithelial spindle cell differentiation is regarded as important [8]. However, information about these cell types is quite few. AME of the breast is usually a solid neoplasm. Nevertheless, cystic changes of AME is also known [4, 6]. Presently, we report a unique case of malignant AME which was located in large cystic structures. Significance and possible histogenesis are discussed here.

3. Case Presentation

A 51-year-old woman visited our hospital with palpable mass in the breast. Physical examination revealed an irregular mass 6cm in diameter in the upper-outer quadrant of the right breast. Doppler ultrasound examination proved cystic structures with different size and a solid mass with size of 2cm connecting to the thick wall of one of cysts, suggesting an intracystic neoplasm. Furthermore, CT examination indicated that the lesion was a congregation of cysts with different size up to 4cm (Figure 1). However, core needle biopsy for the cyst wall, no neoplastic changes were found.

The patient underwent lumpectomy 7 days after the hospital admission. Cut surface of lumpectomy specimen 5cm in diameter was somewhat similar to that of intraductal papilloma (Figure 2). Histologically, luminal surface of the cysts were mostly connective tissues. In some places, atrophic cuboidal cells or flat epithelial cells were present. The solid nodular tissue in the large cyst at the specimen revealed the presence of many tubules lined by an attenuated lining of dark epithelial cells with bland nuclei surrounded layer of clear cells (Figure 3). Cellular atypia was not obvious in both epithelial and myoepithelial cells with low degree of nuclear pleomorphism. In some places, boss types cells existed as randomized population without forming tubule structures. Immunohistochemical activity of the inner epithelial cells with dark cytoplasm was CK 7(+), CAM 5.2(+), CK5/6(partly +), CD10(-), p63(-), S100(-), αSMA(-) (Figure 4). Activity of the myoepithelial cells with clear cytoplasm was p63(+), CK14(+), CD10(+), CK5/6(partly +), CK7(weakly +), S-100(+), αSMA(+) (Figure 5). In some place of the neoplasm, was present cluster of the epithelial cells with advanced cellular and nuclear atypia with increased mitosis (Figure 6). In the present case, spindle cell proliferation emanating from the myoepithelial component was present. They had clear cellular atypia with increased cellular mitosis, enlarged nuclei and nucleoli. Some part of the spindle cell proliferation was sarcomatous (Figure 7). Imunohistochemically, they were p63(+), CK14(+), CD10(+) (Figure 8). These pathological findings suggest to indicate that the neoplasm was malignant AME. For the present patient, round shape shadow in 12mm diameter emerged in the right lower lung in one year after the lumpectomy. Pathological examination after the partial pulmonary lobectomy, the lesion was recognized as almost same morphology as the neoplasm of the breast, and the lesion was diagnosed as metastasis from the malignant AME of the breast. The patient is now under observation.

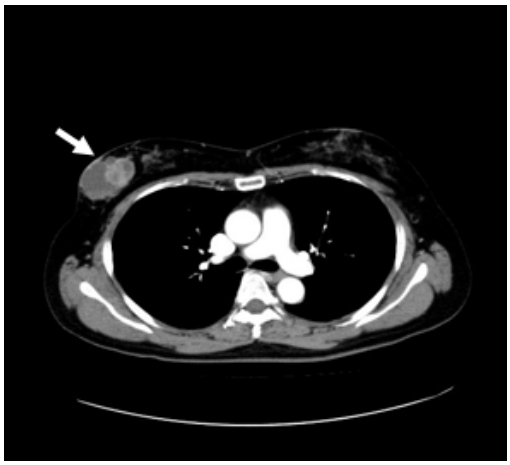

Figure 1: CT examination of the breast lump displaying cystic structure (arrow)

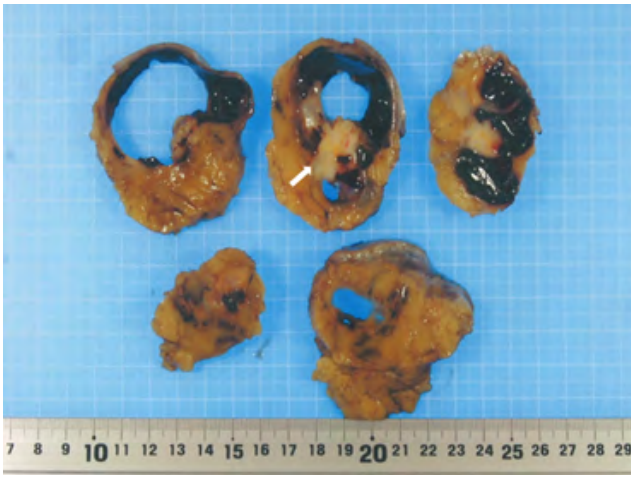

Figure 2: Cut surface of the lump. Arrow indicates nodular lesion of the neoplasm

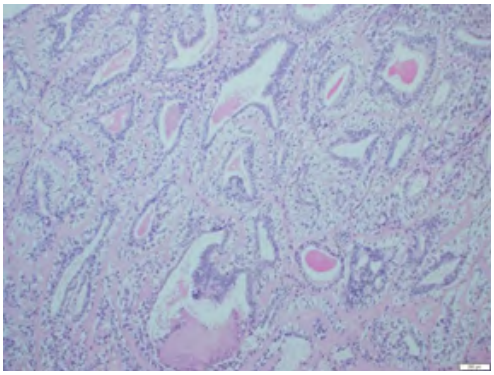

Figure 3: Histology of the malignant AME. Biphasic proliferation of inner epithelial cells and outer myoepithelial cells is shown (HE).

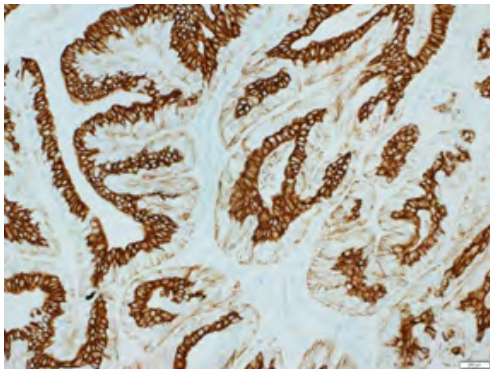

Figure 4: Inner epithelial cells are clearly displayed (CK7).

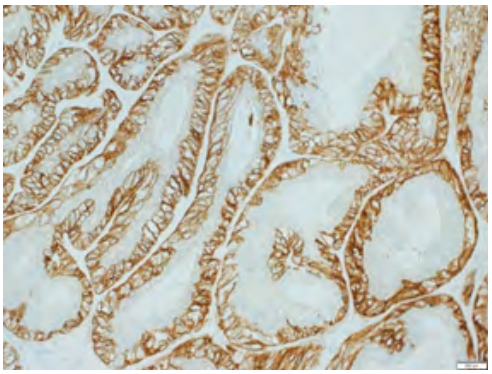

Figure 5: Outer myoepithelial cells are displayed (CD10).

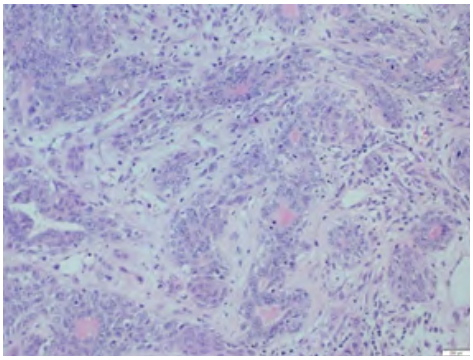

Figure 6: Atypical cell population of the dark epithelial cells (HE)

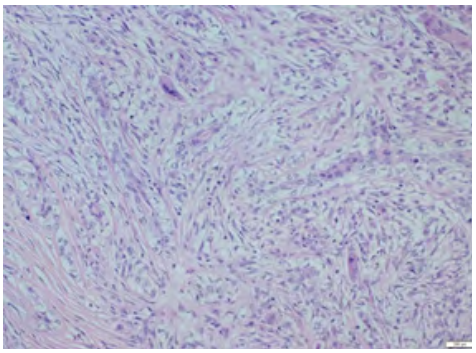

Figure 7: Atypical spindle cell proliferation (myoepithelium origin)(HE)

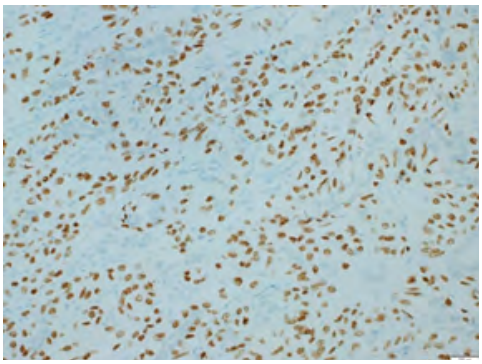

Figure 8: Spindle cell population (p63)

4. Discussion

In contrast to the statement of WHO, recent studies have reported that regional and distant metastasis may occur in about 50% of pure myoepithelial carcinomas [8]. High incidence of metastases of AME to the lung, liver, kidney, heart and skin suggest the high degree potential of malignancy of this neoplasm. In fact, distant metastasis (lung) occurred in one year after the lumpectomy of this case suggesting again the property of malignancy of AME. Morphology of this case including atypical epithelial cells and myoepithelial cells taking spindle cell proliferation support malignant transformation of AME. Recent study suggests that amplification of c-MYC gene is a common pathway to malignant transformation of AME [9]. Allelic imbalance and microsatellite instability of adenomyoepithelioma have also been noted [10]. Such evidence may be also concerned with the transformation. However, detail of the process of malignant transformation is not clear.

In the metastatic lesion of lung, atypical spindle cell proliferation emanating from myoepithelial component was not confirmed, although the biphasic proliferation of neoplastic epithelial and myoepithelial cells was kept as the primary AME. This implies that spindle cell proliferation was not always major component of the malignant AME, although there is a report showing that predominant myoepithelial spindle cell differentiation is important for distant metastasis of AME [8].

Presently, AME was located as a solid mass in the large cystic structures with thick walls. It is known that AME has cystic changes [4, 6]. Therefore, as one of possibility, cystic changes of the malignant AME developed the large cystic structures. However, in this case of malignant AME, the size of solid mass for the neoplasm was quite small and necrotic changes were seen only sporadically. Accordingly, we surmised that immature type of AME emerged in the preexisting cystic structures such as ductal ectasia, hemorrhagic cysts or cyst derived from focal infarction, then, malignant AME grew up by transformation of AME. Furthermore, it might be possible that some stem cells with growing potential toward AME, hiding in the cyst wall is an initial step for the development of AME.

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