G Chir Vol. 39 - n. 4 - pp. 255-257
July-August 2018

clinical practice

Adenomyoepithelioma of the breast: case report and literature review

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SUMMARY: **Adenomyoepithelioma of the breast: case report and literature review**

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Adenomyoepithelioma are uncommon tumors. The majority of them occur in women in the fifth and sixth decades who usually present with a self-palpated, solitary breast mass or a lesion identified on mammography. We report the case of adenomyoepithelioma of the breast with malignant transformation of both myoepitheliel and epithelial components diagnosed as malignancy during the preoperative stage in a seventy-six year old woman.

KEY WORDS: Breast cancer - Breast adenomyoepithelioma.

Introduction

Adenomyoepithelioma (AME) is an uncommon tumor and around twenty malignant adenomyoepitheliomas have been reported in literature (1). The majority of them occur in women in the fifth and sixth decades who usually present with a self-palpated, solitary breast mass or a lesion identified on mammography (2, 3). Less common presentations include pain and nipple discharge. The AME is usually a biphasic tumor, either benign or with low potential of malignancy (4). Morphological features of malignancy that could predict the potential for local recurrence and/or metastasis are not well-established but include necrosis, cellular pleomorphism, overgrowth of myoepithelium or epithelium, invasion and high mitotic activity (1, 5). The World Health Organization (WHO) categorizes adenomyoepithelioma into benign and malignant forms, in which the epithelial component and/or the myoepithelial component shows malignant transformation (6). We report a case of adenomyoepithelioma of the breast with malignant transformation of both myoepitheliel and epithelial components diagnosed as malignancy during the preoperative stage.

Case report

A 76-year-old patient with a clinical history of arterial hypertension and endometrial cancer was admitted in our department. In March 2017, the patient developed pain and a mass in the left mammary region. A bilateral mammography and mammary ultrasound (US) were performed in which was identified a nodular formation with an irregular shape of about 3cm in the upper left quadrant. An US-guided core biopsy was performed and the histological finding was of a poorly differentiated ductal invasive carcinoma (B5 (b) IC G3 ER: neg, PgR: neg, MIB-1: 40%, P53: 70%, C-ERBb 2 / NEU: neg) with extensive necrosis. The patient underwent an upper left quadrantectomy surgery and a frozen section of

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axillary sentinel lymph node, which showed no evidence of malignancyin the definitive histological examination. The patient was dismissed in the first postoperative day. For the clinical staging, the patient underwent a chest X-ray, upper abdomen ultrasound and abone scan. All the examinations were negative. The patient refused to undergo any adjuvant treatment and at one year of follow up shows no sign of tumor recurrence.

Pathological findings

Gross examination of the surgical specimen showed the presence of a solid, hard, well-circumscribed mass with poltaceous areas and irregular margins. The mass was 3.8cms in maximum diameter and had a yellowish color with tan areas.

At microscopic examination, the tumor was circumscribed but lacked a fibrous capsule and was composed in part of tubules and in part of trabeculae separated by strand of thick basement membrane lined by epithelial eosinophilic and myoepithelial clear cells. Both cellular components showed marked atypia with nuclear enlargement and hyperchromasia and prominent nucleoli (Figure 1 A, B). Ten to 12 mitoses for 10 high-power fields were detected both in epithelial and in myopepithelial cells. Areas of coagulation necrosis were present. Immunohistochemical characterization confirmed the biphasic nature of the tumor since myopithelial cells were positive for p63, smooth muscle actin, S-100 and CK34betaE12 and focally positive for CD117, while epithelial cells were negative for these antigens (Figure 1 C, D). Thus, a diagnosis of malignant AME was made.

Discussion

Adenomyoepithelioma lesions are characterized by the presence of both the glandular and myoep-

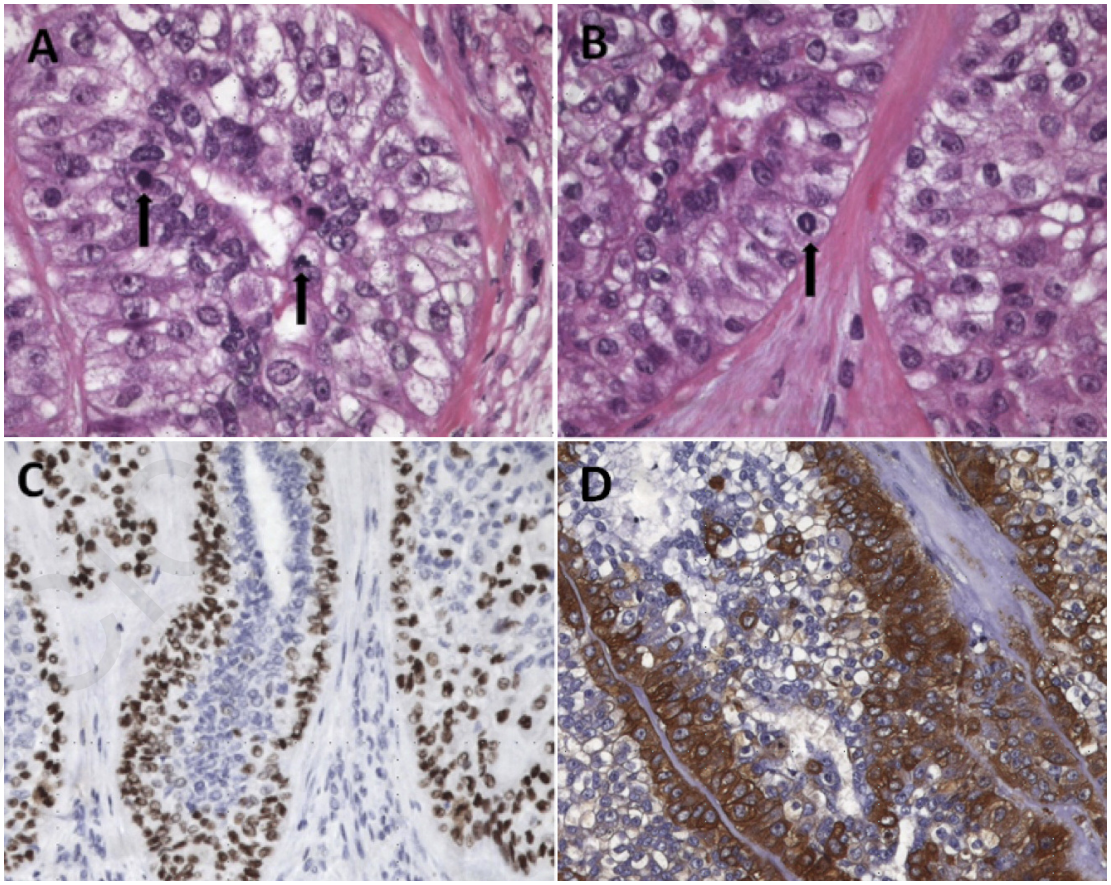

Figure 1 A, B, C, D - A, B) Atypical mitosis (arrows) are seen both in epithelial and in the myoepithelial component (H&E 400X). C) Immunohistochemical stain for p63 is positive in myoepithelial cells (400X). D) Immunohistochemical stain for smmoth muscle actin in positive in myoepithelial cells (400X).

ithelial elements. They belong to a spectrum of myoepithelial lesions which include myoepitheliosis, adenomyoepithelial adenosis, and adenomyoepithelioma lesions. Tavassoli described three variants (spindle, tubule, and lobulated) with a different spread characteristics (4). AME lesions can be considered either benign or malignant based on cytological features and mitotic activity, malignant adenomyoepithelioma is an entity in which either epithelial or myoepithelial components become malignant (7). Due to the capacity of AME with both epithelial and myoepithelial malignant transformation to metastasize, this entity should preferably be treated by wide local excision with appropriate margins (1). The median time to recurrence was reported of 2.3 years [0.7- 5.7 years], with the majority of them in patients undergone excisional biopsy alone (8). A recent meta-analysis showed that the presence of metastases was described in patients with a tumor of ≥ about 1.6 cm and spread mainly via the blood system (1). However, there are no data about sentinel lymph node removal and/or lymphadenectomy and the adjuvant radiotherapy and/or chemotherapy (1). Here we report a case with no sign of local and systemic recurrency besides the disallowance of the patient to any adjuvant therapy at one year of diagnoses.

At least of our knowledge there are few cases reported in literature. Xu J. et al. reported two cases of adenomyoepithelioma of the breast who underwent complete surgical excision that have been followed up for 5-8 years, and both remained free from local recurrence and distant metastases (9). A case of 60 years old woman who underwent surgery treatment and adjuvant radiotherapy was reported by Petrozza V. et al. (1). Kamei M. et al. presented a case of AME coexisting with ductal cell carcinoma in situ with bloody nipple discharge underlying the possibility of the coexistence of breast cancer when encountering such cases (10). In our case symptoms were pain and mass in the mammary region without any bloody nipple discharge.

Conclusion

Malignant adenomyoepithelioma is an uncommon tumor which should be considered in the differential diagnosis of the other solid lesions of the breast. Due to the rarity of this disease, there are a lot of problematic topics, like establishing the diagnosis, managing the optimal therapy and predicting the outcome.

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