KAFKAS TIP BİLİMLERİ DERGİSİ
Kafkas Journal of Medical Sciences
Kafkas J Med Sci 2016; 6(3):199–201 • doi: 10.5505/kjms.2016.08870

OLGU SUNUMU / *CASE REPORT*

Pleomorphic Adenoma of the Breast: A Rare Case Report

Memenin Pleomorfik Adenomu: Nadir Bir Olgu Sunumu

Beyhan Mollamehmetoğlu[1], Burcu Kemal Okatan[1], Çiğdem Şiviloğlu[1], Neslihan Kayaoğlu[2]

[1]Kanuni Education and Research Hospital, Department of Pathology, Trabzon, Turkey; [2]Kanuni Education and Research Hospital, Department of Biochemistry, Trabzon, Turkey

ABSTRACT

Pleomorphic adenoma, also known as mixed tumor, is the most common tumor type in the salivary glands: Most commonly the parotid gland. Pleomophic adenomas may also occur at other sites including the larinx, paranasal sinuse, palete, nasal septum, vulva, and skin (chondroid syringoma).

Pleomorphic adenoma of the breast is rare, and to date only 74 cases have been documented in the word literature. A 53-year-old woman was referred to a phyician with 3 month history of a painless palpable breast mass. A mammogram performed showed a 13 mm, lobulated high-density mass in the immediately below the left nipple. The neoplasm consisted of epithelial, stellate, and spindle cells in pale blue myxoid and hyalinized stroma. The aim of this report is to reitrate and alert the clinicians about rare occurrences including pleomorphic adenomas in the breast. Particularly in the presence of suspicious clinicoradiologic findings, an awareness of these benign lesions will help render an accurate diagnosis and prevent unnecessarily aggressive surgery.

Key words: *pleomorphic adenoma; breast; immunohistrochemistry*

ÖZET

Benign miks tümör olarak bilinen pleomorfik adenom, tükrük bezlerinin sıklıklada parotis bezinin en çok görünen tümör tipidir. Tipik olarak ağrısız, sabit benign kitledir. Ayrıca larinks, paranazal sinus, palatina, nazal septum, vulva ve deride (kondroid siringoma) oluşabilir. Geniş eksize edilmezse pleomorfik adenom nüks edebilir.

Pleomorfik adenom memede nadirdir ve dünya literatüründe bu güne kadar sadece 74 olgu bildirilmiştir. 53 yaşında kadın hasta üç aydır fark edilen ağrısız meme kitlesi ile doktora başvurdu. Mammogramda sol meme başının hemen altında yüksek dansiteli lobule 13 mm kitle izlendi. Neoplazm, açık mavi miksoid ve hyalinize stromada epiteliyal, stellat ve iğsi hücrelerden oluşmaktadır. Bu sunumun amacı bu nadir meme kitlesi hakkında klinisyenleri uyarmak ve yeniden hatırlatmaktır. Özellikle şüpheli klinikoradyolojik bulgular varsa bu benign lezyonun farkındalığı doğru tanıyı koymada ve gereksiz aşırı cerrahiyi önlemede yardımcı olacaktır.

Anahtar kelimeler: *pleomorfik adenom; meme; immunohistrochemistry*

Uzm. Dr. Beyhan Mollamehmetoğlu, Trabzon, Türkiye
Tel. 0532 770 90 47 Email. bmollamehmetoglu@yahoo.com
Geliş Tarihi: 30.03.2015 • Kabul Tarihi: 12.09.2016

Introduction

Pleomorphic adenoma, also known as mixed tumor, is the most common tumor type in the salivary glands: Most commonly the parotid gland. It is a benign tumor which typically presents as a painless, persistent mass[1,2,3]. Pleomophic adenomas may also occur at other sites including the larinx, paranasal sinuse palete, nasal septum, vulva, and skin (chondroid syringoma)[2]. Pleomorphic adenoma of the breast is rare, and to date only 74 cases have been documented in the world literature[1].

The aim of this report is to re-iterate and alert the clinicians about rare occurrences including pleomorphic adenomas in the breast. Particularly in the presence of suspicious clinicoradiologic findings, an awareness of these benign lesions will help render an accurate diagnosis and prevent unnecessarily aggressive surgery.

Case Report

A 53-year-old woman was referred to a phyician with 3 months history of a painless palpable breast mass. A ultrasound performed showed a 13mm , hypoechoic mass in the immediately below the left nipple. The physical examination was remarkable for a 13 mm round to oval mobile mass present at subareolar region within the left breast.The mass was described as firm in consistency. No other discrete mass was present. There was no nipple retraction or discharge. No axillar or supraclavicular lympadenopaty was noted. The preoperative clinical and radiological impression was that of an intraductal papillomatous lesion. The mass was completely excied. Gross examination showed a single fragment of firm pale white tissue measuring 3×2.5×2.5 cm, which was serially sectioned to show a 13-mm circumscribed solitary mass.

Histologic examination showed a circumscribed neoplasm (Fig. 1) surrounded by fatty breast tissue. The neoplasm consisted of epithelial, stellate, and spindle cells in pale blue myxoid and hyalinized stroma (Fig. 2). The epithelial component consisted of relatively uniform appearing cells having round to slightly ovoid hyperchromatic nuclei arranged in tubules, cords, and small nested aggregates. The stellate and spindle-shaped cell were seen in myxohyaline stroma (Fig. 3). No condroid metaplasia was seen. A duct ectasia (periductal mastitis) was also identified immediately adjacent to the neoplasm. The stroma stained focaly with musicarmine and alcian blue (at pH 2.5).

Results of immunohistochemical staining were consistent with pleomorphic adenoma. The myoepithelial-type cells stained positively with S-100 protein (Fig. 4), vimentin, smooth muscle actin, and glial fibrillary acidic protein (GFAP). Epithelial cells stained positivity for keratin AE1/AE3, epithelial membrane antigen (EMA), carcinoembriyonic antigen (CEA, focal).

Discussion

A benign mixed tumor of the breast (PAB) is an extremely rare benign neoplasm, accounting for 74 cases in the world literature[1,3]. It has been reported in other, less common sites such as nasal septum, paranasal sinuses, larynx, palate and vulva[2]. It also occurs in the skin, where it is more commonly known as chondroid syringoma. PAB was first reported in 1906 by Lecene[5], who described a case with cartilaginous and osseous

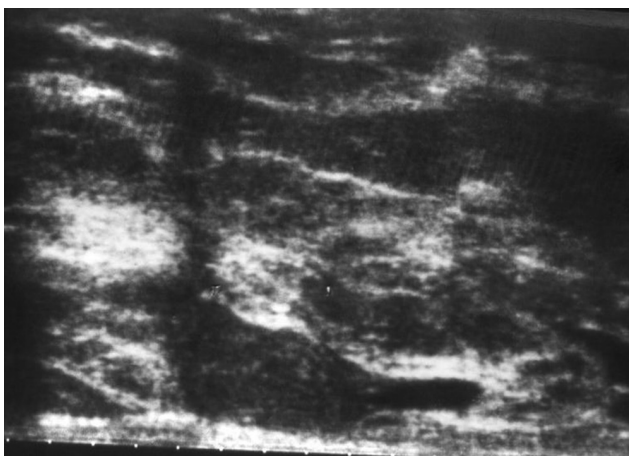

Figure 1. *Ultrasonographic examination showing solid hypoechoic mass.*

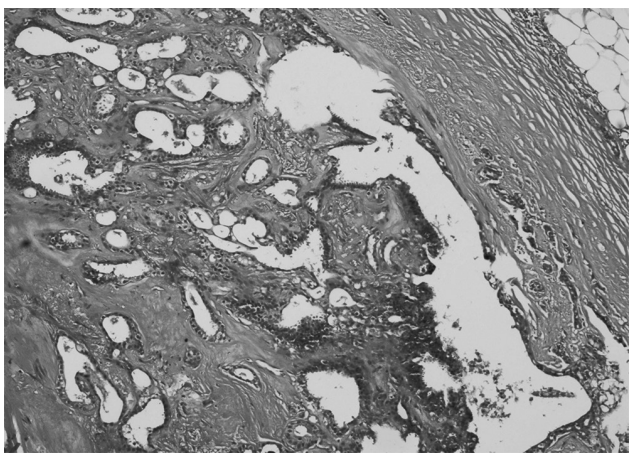

Figure 2. *The tumor has well circumscribed border. Epithelial and myoepithelial cells can be easily distinguished (hematoxylin and eosin ×100).*

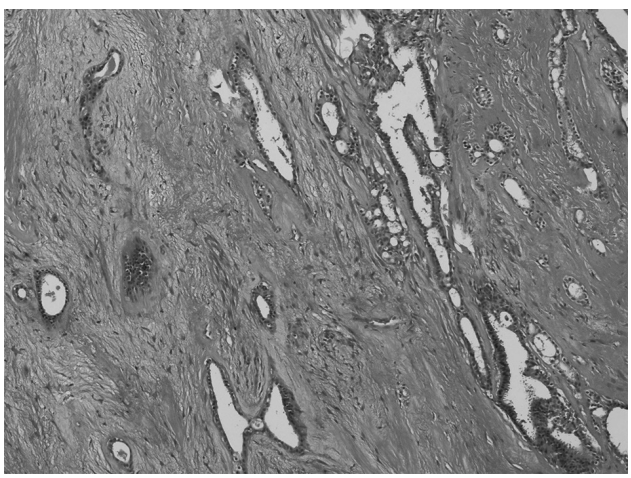

Figure 3. *Epithelial structures with glanduler pattern are embedded in loose, chondromyxoid to fibrous stroma (hematoxylin and eosin ×200).*

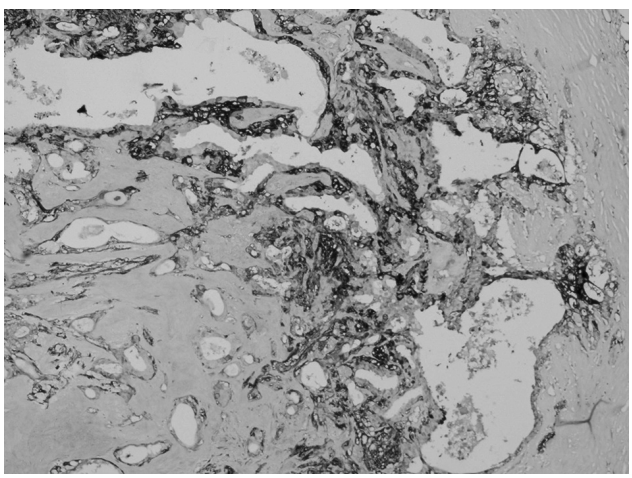

Figure 4. *Immunostain for S100 protein. Myoepithelial cells of pleomorphic adenoma show positive cytoplasmic immunoreactivity (original magnification ×100).*

metaplasia. Patiens can range in age from 23 to 85 years. Tumor size range from 0.6 to 20 cm[4]. PAB has the tendency to occur in the periareolar region (like our case), and this may suggest that it originates from the large duct. It has led to speculation that PABs may intraductal papillomas, adenomyoepitheliomas that have areas of osseous and chondroid stroma rather then a saperate kind of neoplasm. Other authors believe that this kind of tumor is separate entity. Intraductal papillomas form distinct papillary structures, and adenomyoepitheliomas show a predominant proliferation of polygonal myoepithelial cells with clear cytoplasm, which may surround gland-forming epithelial cells. Moreover, the presence chondroid and osseous metaplasia are rare component of intraductal papillomas and adenomyoepitheliomas[1,3].

Morphologically, PAB shows a remarkable degree of morphologic diversity. It is composed of a mixture of epithelial, myoepithelial, and stromal or mesenchymal elements. Epithelial cells appear cuboidal to columnar with bland cytologic nuclear features and low mitotic activity. They are arranged as tubular structures, islands, cords, duct-like structures, sheeds and may exhibit apocrine differantiation. Myoepithelial cells appear polygonal, plasmacytoid, fusiform, or stellate with small nuclei and clear to eosinophilic cytoplasm. The mesenchymal component can be myxoid, mucoid, cartilaginous. Stromal musin can be demostrated with colloidal iron and mucicarmine stains[2,3,5]. By immunohistrochemistry, the inner ductal cells are positive for cytokeratin, carcinoembriyonic antigen, and epithelial membrane antigen. Myoepithelial cells are strongly positive for vimentin, muscle-spesific actin, calponin, CD10,GFAB and cytokeratin. S100 protein is typically more intense in myoepithelial cells[6].

The differential diagnosis of PAB includes metaplastic carcinoma with cartilaginous and osseous metaplasia. However, metaplastic carcinoma are characterized by the presence of poorly differentiated carcinoma admixed with atypical or malign mesenchymal elements. Colloid (mucinous) carcinoms are another differential diagnosis so this tumor is cytologically bland and has abundant myxoid stroma. Alcian blue staining with concomitant hyaluronidase treatment could serve as a simple stain to help differentiate these two antities[3]. Another differential diagnosis is intraductal papilloma with myxochondroid stroma. Intraductal papilloma can be distinguished by their lack of proliferatifve myoepithelial elements,which is characteristic of PAB. In all reported cases, the benign nature of PAB is exemplified by the absence of metastases in all reported cases. Extensive surgical excision with clear margin is the single therapeutic option.

In conclusion, pleomorphic adenoma is rare benign tumor of the breast and may be confused with a number of benign and malignant tumors, particularly in the presence of suspicious clinicoradiologic findings. Careful paraffin sections, immunohistochemical studies and special stains should be performed to facilitate diagnosis in especially difficult cases and prevent unnecessarily overaggressive surgery.

References

1. Khamechian T, Alizargar J, and Mazoochi T. Reporting a rare case of pleomorphic adenoma of the breast. Case Rep Med 2014;2014:387183.
2. Su A, Apple SK, Moatamed NA. Pleomophic adenoma of the vulva, clinical reminder of a rare occurrence. Rare Tumor 2012;4: e16.
3. Reid -Nicholson M, Bleiweiss I, Pace B, Azueta V, Jaffer S. Pleomophic adenoma of the breast. A case report and distinction from mucinous carcinoma. Arch Pathol Lab Med 2003;127:474–77.
4. Sato K, Ueda Y, Shimasaki M, at al. Pleomorphic adenoma (benign mixed tumor) of the breast: a case report and review of the literature. Pathol Res Pract 2005;201:333–9.
5. Chen KT. Pleomorphic adenoma of the breast. Am J Clin Pathol 1990;93:792–94.
6. Diaz NM, McDivitt RW, Wick MR. Pleomorphic adenoma of the breast: a clinicopathologic and immunohistochemical study of 10 cases. Hum Pathol 1991;22:1206–14.