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內文：

一. Introduction

Pleomorphic adenoma(PA) is well known to be a

1. benign salivary gland tumor
2. malignancy arises in PA, it is usually carcinoma.

Myoepithelial carcinoma

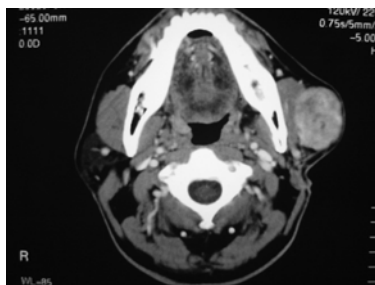
1. extremely rare in salivary gland
2. less than 1% of all salivary gland tumors.
3. develop de novo or may appear in a pre-existing PA.

We herein report the case of a **myoepithelial carcinoma with a high grade of malignancy arising from a recurrent pleomorphic adenoma**. The histological findings of both the initial and the recurrent neoplasm are described in detail.

二. Case report

A 53-year-old male

1. swelling of the left parotid region in 2003.
2. a superficial left parotidectomy in 2001 for the treatment of a pleomorphic adenoma.

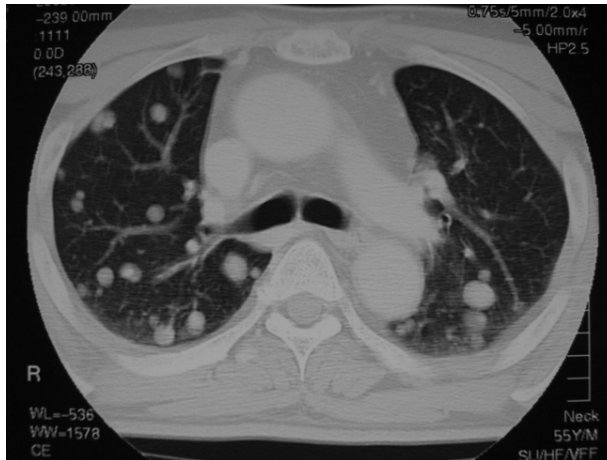


3. Palpation: hard, 6 cm .
4. No facial nerve palsy

A computed tomographic (CT) study showed a **large tumor in the left parotid gland**. **No lymph node enlargement** was

noted.

5. Fine needle aspiration biopsy: features of pleomorphic adenoma.

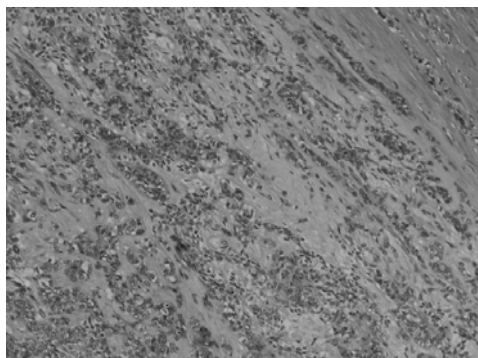


Multiple coin lesions were also found in the lungs on both chest X-ray films and computed tomography

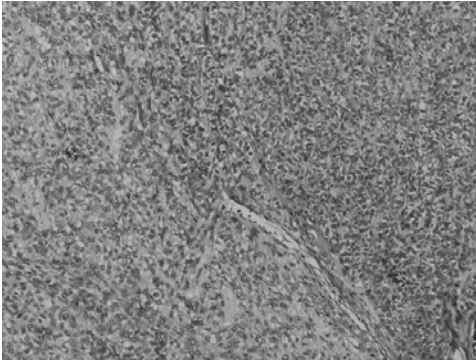
6. A left total parotidectomy was therefore performed, and the capsulized tumor which could be easily separated from the facial nerve. Stenson's duct was resected due to tumor infiltration. In frozen sections, the histopathological diagnosis was a recurrent pleomorphic adenoma with a proliferation of myoepithelial cells.
7. Partial lung resection biopsy under thoracoscopy was also performed.
8. The histopathological examination revealed myoepithelial carcinoma of both the parotid gland tumor and the lung tumor, occurring simultaneously with the recurrent pleomorphic adenoma.
9. Using chemotherapy, but died 3 years after the initial operation.

三. Pathological findings

1. Both the initial tumor and the recurrent tumor were reviewed.



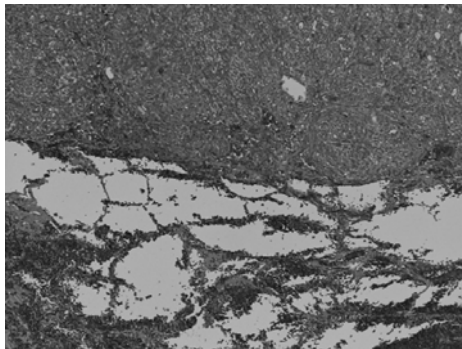
The initial tumor showed a mixture of myxoid, solid, and chondroid stroma. Ductular formations of round cells were found. No cytological atypia, mitosis, or necrosis was seen.



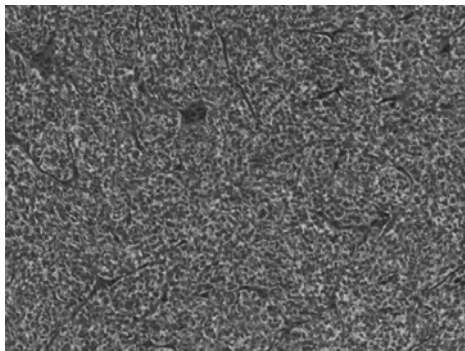
In a certain area of the initial tumor, a **hypercellularity of the myoepithelial** tissue was found

Although the resection **margin was free from the tumor**, some tumor cells had invaded the capsule.

Consequently, the neoplasm was diagnosed to be **pleomorphic adenoma with a predominance of myoepithelial cells**.



The recurrent tumor showed the monotonous growth of **atypical epithelial round cells** with any **abundant mitosis**, as well as neither **necrosis** nor hemorrhage.



The parotid gland and the lung tumor were very similar except for the presence of tubular formations in the lung tumor, which consisted of myoepithelial cells. Some multinucleated cells were also present.

2. The immunohistochemical markers

- Anticytokeratin: positive
- Anti- α smooth muscle actin: partially positive
- Anti vimentin: partially positive,
- Anti s-100 protein: diffusely positive
- Alcian-blue: negative.

3. Final diagnosis: myoepithelial carcinoma with a high grade of malignancy arising from a recurrent pleomorphic adenoma.

4. A total parotidectomy performing (controversial due to chest CT image, but p't request)

四. Discussion

1. **De novo formation** is an indication of a **high-grade malignancy** and a **secondary formation** from a pleomorphic **low-grade malignancy**
2. The prognostic implications of the histogenesis of the myoepithelial carcinoma therefore remain controversial.
3. The **first extracted tumor(pleomorphic adenoma)** showed a special feature in the present case. It was different from common mixed tumors in that a **hypercellularity of the myoepithelial cells** was found in a certain area. The **tumor infiltrated the capsule** of the tumor, but not the extracapsular area, and it **lacked cytological atypia, mitosis, and necrosis**.
4. In the literature, a **myoepithelial proliferation** has been identified as a possible **predictor** of an **aggressive** clinical behavior of the benign pleomorphic adenoma. **Cresson** et al. stated that **aneuploidy**(abnormal number of chromosomes) in a **DNA flow cytometry** of the cells from the tumor might reflect the malignant potential of a pleomorphic adenoma.
5. It remains **controversial** whether this neoplasm was classified as a **myoepithelial carcinoma** or a **carcinoma ex pleomorphic adenoma** in the classification by the WHO in 2005.
6. Histopathologically, undifferentiated carcinoma, adenocarcinoma, and squamous cell carcinoma are mainly observed as the malignant components of a carcinoma ex pleomorphic adenoma. A **myoepithelial carcinoma as a malignant component is rare**.
7. In cases of **carcinoma ex pleomorphic adenoma**, **both carcinoma and adenoma components** were supposed to exist in the primary lesion. However it is often difficult to demonstrate a pre-existing pleomorphic adenoma because the malignant component may overgrow the other component.
8. In the present case, there was no evidence of the existence of adenoma in the recurrent tumor despite the examination of multiple sections. Nevertheless, the presence of **tubular formations in the lung tumor** might have been a vestige of the pleomorphic adenoma. **Namely the present neoplasm was supposed to be a “myoepithelial carcinoma ex pleomorphic adenoma.”**

9. Pleomorphic adenoma with a hypercellularity of the myoepithelial cells should therefore be treated as a pre-malignant neoplasm.

Question

題號	題目
1	What is the most occurrence of Benign Tumor of the salivary gland? (A) Pleomorphic adenoma (B) Warthin tumor (C) Oxyphilic cell adenoma (D) Myoepithelioma
答案 (A)	出處 : Oral and maxillofacial pathology, 2nd
題號	題目
2	Which is the most occurrence of the malignant tumor of the salivary gland? (A) Mucoepidermoid carcinoma (B) Acinic cell carcinoma (C) Carcinoma ex pleomorphic adenoma (D) Adenoid cystic carcinoma
答案 (A)	出處 : Oral and maxillofacial pathology, 2nd