CASE REPORT

Unicystic variant of low grade mucoepidermoid carcinoma: A rare case report

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Abstract

The most common malignant neoplasm of salivary gland origin is mucoepidermoid carcinoma (MEC), and it comprises of about 5-10% of all salivary gland neoplasm. It is mostly seen in major salivary glands (54%) and two third of them occurs in the parotid gland, and 46% is seen in minor salivary glands. Histo-morphological complexity of MEC has a constellation of varying amount of epidermoid cells, goblet cells, intermediate cells and mucocytes. It is graded histopathologically based on the relative proportion of cell type, cystic spaces, degree of tumor invasion, anaplasia, mitotic rates and presence or absence of necrosis into low-grade, Intermediate grade and high-grade. In the literature, several variants of MEC such as conventional, clear cell, warthin-like, sclerosing and unicystic variant have been reported. The aim of the current case report is to present a low-grade variant of unicystic MEC which is rare, but a distinctive tumor affecting the salivary gland. Till date, to the best of our knowledge only one case has been reported.

Keywords

Mucoepidermoid carcinoma, salivary neoplasm, unicystic

Introduction

One of the most common neoplasms of the salivary gland origin is mucoepidermoid carcinoma (MEC) and accounts for about 5-10% of all salivary gland origin. Stewart et al. in 1945 described this lesion at first and is thought to arise from pluri-potent reserve cells of the excretory duct of salivary gland that have potential to differentiate into squamous, columnar and mucus cells.[1,2]

Exposure to ionizing radiation is considered to be one of the etiological factors for MEC, and this neoplasm is commonly seen in children and adolescent. Histologically it is composed of varying amounts of epithelial cells, intermediate cells and mucus cells (often seen lining the micro cysts). The combination of cellular element in varying proportions can lead to complex histological pattern leading to diagnostic challenges.[3,4]

MEC is graded into three categories such as low-grade (tumor exhibiting greater than 50% of mucus elements); intermediate grade (about 10-50% of mucus elements is present) and high-grade (10% or less of mucous elements is present). The histopathological grading is used as a prognostic indicator, wherein the low-grade MEC has a better 5 year survival rate with 92-100% compared to high-grade MEC with 0-43% survival rate.[5,6]

Here, we report a unique case of unicystic variant of low-grade MEC in an adult patient and further

Figure 1: (a) Cyst exposed during excision, (b) enmass excised cystic lesion, (c) cut section revealed smooth and glistening lining
insist to consider this rare entity in the differential diagnosis of cystic lesions of the parotid gland and of the oral cavity.

**Case Report**

A 44-year-old woman reported with a painless swelling in the right pre-auricular region since 1 year. Swelling was gradually increasing in size and measured approximately 4 cm × 3 cm in size with no signs of discharge. On clinical examination, swelling was soft in consistency, mobile, nontender and not fixed to underlying structures nor adherent to the overlying skin. Facial nerve was intact with no cervical lymphadenopathy. A provisional diagnosis of pleomorphic adenoma was given.

Fine needle aspiration revealed a deep yellow viscous fluid. On surgical removal, a thick walled cystic lesion was found and submitted for histopathological diagnosis.

On gross examination, the cyst was round in shape with a smooth surface. Cut section revealed a single large cystic space filled with yellowish viscous fluid [Figures 1a-c].

On microscopic examination cystic cavity was lined with thin epithelium comprising of epidermoid cells, mucus cells and clear cells. The stroma comprised of proliferation of tumor cells in small islands, groups and glandular pattern. Tumor cells were round and oval in shape with vesicular nuclei and deep eosinophilic cytoplasm. Mucus cells predominated with numerous microcyst formations throughout the section observed. Special stains such as periodic schiff stain, alcian blue and mucicaramine stained positive for mucus cells [Figures 2a, b and 3a-c]. Taking into account of the above histological findings a diagnosis of low-grade MEC of unicystic variant was given.

**Discussion**

MEC is considered the most common malignant salivary gland tumor and is usually seen in children and adolescent under the age of 20 years. Pain is associated with high-grade MEC’s, and facial nerve paralysis is not seen when parotid gland is involved.[5]

Microscopically, MEC consists of epidermoid cells, mucus cells and intermediate cells arranged in cords, sheets or cystic configuration and are classified as low, intermediate and high-grade. As a result of this cellular heterogeneity and histological composition, MEC presents itself with diverse histological variants such as sclerosing, unicystic, oncocytic, sebaceous, clear cell, goblet cell aggressive, spindle cells and of psammomatous types. Among these types, unicystic MEC seems to be very rare variant. To the best of our knowledge, only one case of unicystic MEC have been reported in the English literature and were classified as low-grade tumor with no evidence of recurrence.[3,7]

In more than 50% of the histologic sections of MEC’s studied by Kofi et al. prominent cystic features were observed. Thus, during a physical examination or on ultrasonography of the cystic lesions of parotid glands, one must not dismiss it as any other benign lesions but rather careful examination and further investigation would be helpful.[8]

The treatment of choice for MEC’s is excision involving superficial or total parotidectomy with facial nerve preservation. If there is clinical evidence of regional metastasis or high tumor-node-metastasis stage, and high histological grade with involvement of regional nodes radical neck dissection should be considered.[9]

**Conclusion**

MEC of the parotid gland is a very common salivary gland neoplasm in adults and usually presents as diverse clinical and histological pattern. Present case emphasizes in considering the unicocular cystic variant of MEC in the differential diagnosis of cystic lesions of the parotid gland as well as adds to the minimal literature available.

![Figure 2](image1.png)  
**Figure 2:** (a) H and E section showing cystic cavity lined by thin epidermoid cells along with mucus cells, (b) Stroma comprising of islands of epidermoid cells and mucus cells (×10)

![Figure 3](image1.png)  
**Figure 3:** Photomicrograph showing sections stained with special stains: (a) Periodic acid-Schiff positive mucin stained as magenta pink in color, (b) mucicaramine positive stained section and (c) Alcian blue positive stained as blue in colour (×20)
References


