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WARTHIN LIKE VARIANT OF MUCOEPIDERMOID CARCINOMA SALIVARY GLAND- A RARE ENTITY

Pathology

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland tumor1. Although the parotid gland is the most common site of involvement, other major salivary glands and the minor salivary glands—most commonly of the palate—also can be involved. The management of mucoepidermoid carcinoma depends on the grade of the tumor and the adequacy of resection. We present the case of a 41-year-old gentleman with no known co-morbidities presented with swelling over right parotid region for 20 years. Superficial parotidectomy was done and the histopathology revealed mucoepidermoid carcinoma - Warthin like variant. Extensive literature search has revealed only a few reported such cases.

KEYWORDS

Case history

A 41-year-old gentleman with no known comorbidities presented with painless swelling over right parotid region for last 20 years. Since last 2 years there was a sudden increase in size of the swelling. Ultrasonographic examination of the parotid region showed a well-defined heteroechoic lesion towards lower end of right parotid gland measuring 3.5x3x2.8cm. CT neck revealed irregular heterogenous mildly enhancing lesion in the right parotid gland. Possibilities suggested were Pleomorphic adenoma, Warthin's tumor, necrotic lymph node and malignant salivary gland tumor. The same was reported as a benign lesion on FNAC. Right superficial parotidectomy was done. Grossly specimen measured 6.5x5x2.3cm, which on cut section showed a circumscribed lesion measuring 3.5x3x2.3cm. Cut surface was grey white with cystic spaces filled with mucoid material.(Fig.A) Periphery showed normal looking parotid gland. Microscopic examination of the H&E slides revealed a fairly circumscribed neoplasm composed of cells arranged as variably sized cystic spaces, solid nests and islands. Three population of cells are noted – intermediate-type cells, mucin producing and squamoid cells with mild to moderate pleomorphism, vesicular nuclei and a few with prominent nucleoli.3-4mitotic figures/hpf noted in mitotically active areas. The cystic spaces are filled with mucinous material and focally lined by mucous cells. Intervening stroma shows fibrosis, foci of sclerosis, dense lymphocytic infiltrate, congested vessels and haemorrhage, Cholesterol crystals and necrotic foci noted. No perineural or Lymphovascular invasion noted. In focal areas the cystic spaces were lined by oncocytic cells with intervening lymphoid stroma. Intracytoplasmic mucin also noted.(Fig.B,C,D,E)

A final diagnosis of mucoepidermoid carcinoma-warthin like variant, intermediate grade was given.

DISCUSSION

The diagnosis and management of salivary gland neoplasms are mainly based on the histopathological entity and the grade of the neoplasm[2]. Benign tumors are usually managed by surgery alone, while malignant tumors require surgery with additional adjuvant therapy[2]. Warthin tumor (WT) and Mucoepidermoid carcinoma (MEC) are two histologically distinct salivary gland neoplasms. Warthin's tumor or adenolymphoma is the second most common benign tumor of the salivary glands seen in the elderly population and commonly occurs in the superficial lobe of the parotid gland as a slow-growing indolent mass. Histologically, Warthin's tumor has morphologic characteristics consisting of (i) cystic structures lined by bilayered oncocytic and basaloid epithelium with papillary projections; and (ii) underlying lymphoid stroma. In contrast, MEC is the most common malignant salivary gland tumor, which is histologically composed of a mixture of mucous cells, intermediate cells, and squamous-like or epidermoid cells. MEC and WT are usually not considered in the differential diagnosis of a single lesion because they have distinct clinical and histological diagnostic criteria. However, a tumor showing the morphology of both mucoepidermoid carcinoma and Warthin's tumor is very unusual, as in our case.

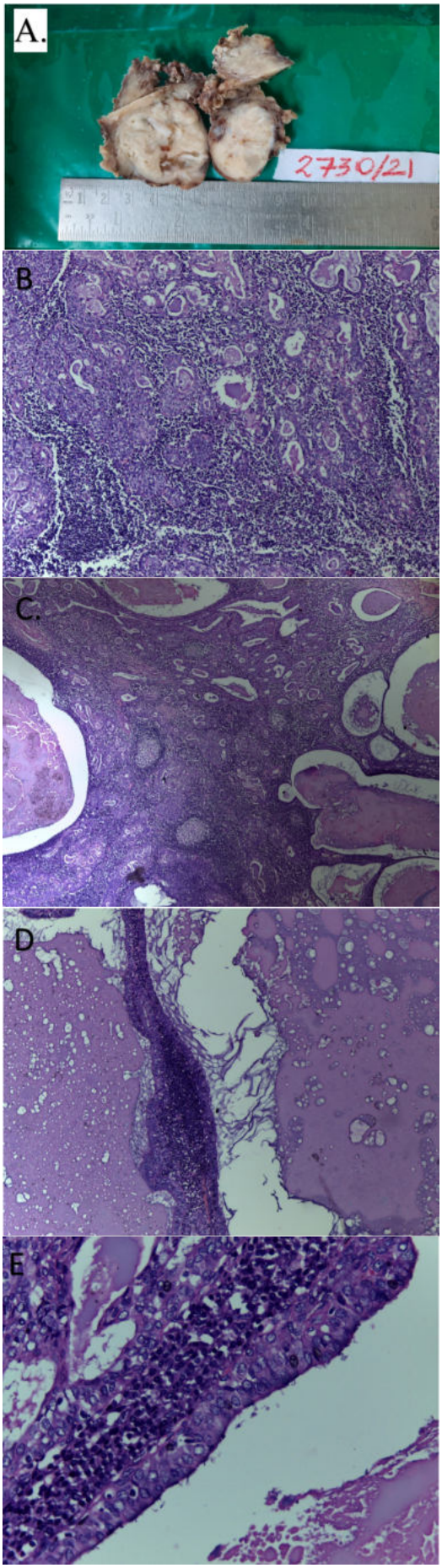
The main differential of such types of lesions are the malignant transformation of WT, metaplastic changes in WT and the Warthin-like variant of MEC. The coexistence of WT and MEC is rare, with only 29 cases reported worldwide.[3-5] Transformation can occur spontaneously or following radiotherapy. Seifert et al.[6] proposed four criteria to support a diagnosis of malignant transformation of WT; namely (i) the presence of a pre-existing benign WT; (ii) the presence of transition zones from benign oncocytic to malignant epithelia; (iii) infiltrating growth in the surrounding lymphoid tissue; and (iv) exclusion of metastases to the lymphoid stromal component of a primary extra salivary tumor.

In 2015, Ishibashi et al.[7] gave the first description of a Warthin-like variant of MEC. A similar histomorphology was also described by Heatley et al[8] in a 17-year-old female presenting with a cystic lesion, which, on initial excision, resembled WT and classical mucoep idermoid morphology in the recurrent tumor. A retrospective review of the initial histology revealed the squamoid appearance and focal presence of mucin, which raised the suspicion of Warthin-like MEC.

Hang et al.[9] described the cytological picture of a Warthin-like MEC as showing dense lymphocytic infiltration and cystic change, which resembled WT on low-power field examination. The tumor epithelium has a metaplastic appearance with squamoid and goblet cells, which could be misinterpreted as intermediate and mucinous cells as in MEC.

The prognosis of MEC depends on the tumor stage and grade, and the adequacy of resection margins.[10] MEC grading is based on the percentage of mucous cells into the low, intermediate, and high grade. Low-grade lesions are usually managed with surgery alone while high-grade lesions or high-risk lesions (with neural invasion, necrosis, mitotic figures, anaplasia), and margin-positive lesions require adjuvant radiotherapy.[5] No case of metastases has been reported in patients with MEC coexisting with WT of the parotid gland treated with margin-free excision. However, one patient presented with local recurrence, whose initial margin status was unknown.[8] The prognosis of Warthin-like MEC is usually considered with better than the classical MEC given its benign component and the lower grade of the MEC component. In our case since all resected margins are free of neoplastic infiltration; the patient is under strict follow up. No further surgical/radiotherapeutic intervention was done.

The presence of the histological picture should raise suspicion regarding this rare and clinically relevant variant. The rarity of this tumor has resulted in the lack of definitive evidence for diagnosis and management. Further research is required before this variant can be clearly defined.



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