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CASE STUDY

MUCOEPIDERMOID CARCINOMA MIMICKING A MUCOUS RETENTION CYST:A DIAGNOSTIC DILEMMA

Dr. Tejashree Pangare, *Dr. Sonal B. Shah, Dr. Pushkar Waknis, Dr. Pratik Suthar, Dr. Lakshmi Shetty and Dr. Mamatha G.S. Reddy

Department of Oral and Maxillofacial Surgery, Dr. D. Y. Patil Vidyapeeth's Dr. D. Y. Patil Dental College and Hospital, Pimpri, Pune -18

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ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common malignant salivary gland neoplasm. It displays a variety of biological behaviour. It affects both the major and the minor salivary glands. The clinical features and biological behaviour of MEC of the minor salivary gland has not been very widely documented. Low grade MEC usually demonstratres a benign nature. Here, we report a case of bilateral soft tissue swellings of the labial mucosa which presented as mucous retention cysts and later diagnosed histopathologically as low grade MEC of the minor salivary gland.

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INTRODUCTION

Mucoepidermoid carcinoma is one of the most common salivary gland malignancies (Kulkarni *et al*., 2014). This neoplasm had once been regarded as a benign lesion and called 'mucoepidermoid tumor' before the revised WHO classification (1990) which settled on the term 'MEC' (Cho *et al*., 1997). MEC is believed to arise from the reserve cells of excretory ducts, and the tumor consists of three cell types: epidermoid cells, mucous cells and poorly differentiated intermediate cells (Ozawa *et al*., 2008). Epithelial salivary gland neoplasms are rare both in adults and children, accounting for <3% of all head and neck tumors (Kulkarni *et al*., 2014). The most common type of malignant salivary gland neoplasm of epithelial (parenchymal) origin in the pediatric-adolescent age group is the MEC. Most of the malignant neoplasms are found in the parotid gland; only a few pediatric and adolescent cases have been well documented in the minor glands (Ritwik *et al*., 2012). Here we are reporting a unique, rare case of bilaterally symmetrical MEC affecting the upper labial mucosa.

Corresponding author: Dr. Sonal B. Shah,
Reader, Department of Oral and Maxillofacial Surgery, Dr. D.Y. Patil Vidyapeeth's , Dr. D. Y. Patil Dental College and Hospital, Pimpri, Pune -18.

CASE REPORT

A 65-year-old male patient reported to our Department of Oral and Maxillofacial Surgery with the chief complaint of bilaterally symmetrical mucosal swellings of the upper labial mucosa since 1 year. He gave a history of cheek bite while having food following which he experienced pain in the upper right and left front region of the lip.

The pain was dull and continuous in nature and aggravated on mastication. One year back, he noticed small swellings on the right and the left side of the upper labial mucosa. He bit the swellings which was followed by a salty discharge and the swellings regressed. They would reappear again in 15 days time and this continued for a year. He had been to a local dentist who clinically diagnosed it as a mucous retention cyst.

The clinical examination extraorally revealed that the swellings appeared as rounded protuberances above the upper lip bilaterally. The skin over the concerned area was normal. On palpation they were soft swellings which were non fluctuant, tender and not fixed to the overlying skin. Intra oral examination revealed a well-defined solitary round soft sessile, tender swellings present in right and the left upper labial mucosa in front of 13, 14 and 23, 24 respectively measuring

1.5 cm × 1.5 cm in its greatest dimension. The overlying mucosa was normal. On palpation, inspectory findings were confirmed. The swelling was tender, soft in consistency, mobile and non-fixed to underlying structures. The swelling was non-fluctuant, non-compressible and non-pulsatile. A panoramic radiograph was done so as to rule out any pathology associated with the teeth and bone.

The clinical presentation was confirmatory of the lesion being a mucous retention cyst. Antibiotics were started and after the pain was reduced, excisional biopsy was done. The lesion was removed in toto. Histopathological examination revealed prominent cystic structures of varying thickness lined by mucous cells and epidermoid cells. Few cystic spaces lumen were filled with pale eosinophilic mucin. Mucous cells were round to ovoid with pale foamy cytoplasm. Adjacent fibrous connective tissue showed few clusters of normal salivary gland acini with focal aggregates of chronic inflammatory cell infiltrate. All these features were suggestive of the lesion being a low grade variant of MEC.

Fig.1. Pre-Operative Extraoral View

Fig.2 Pre-Operative Intraoral View

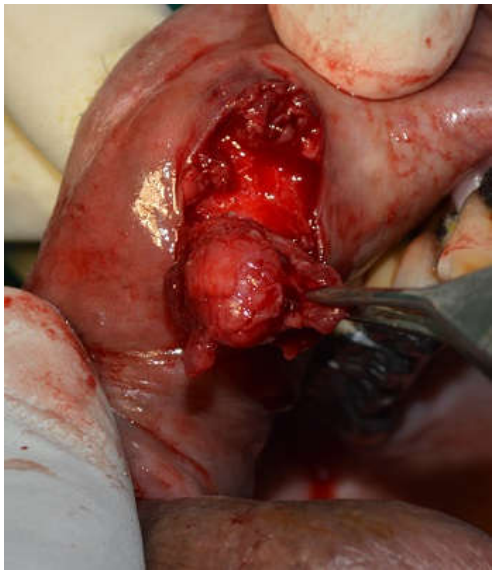
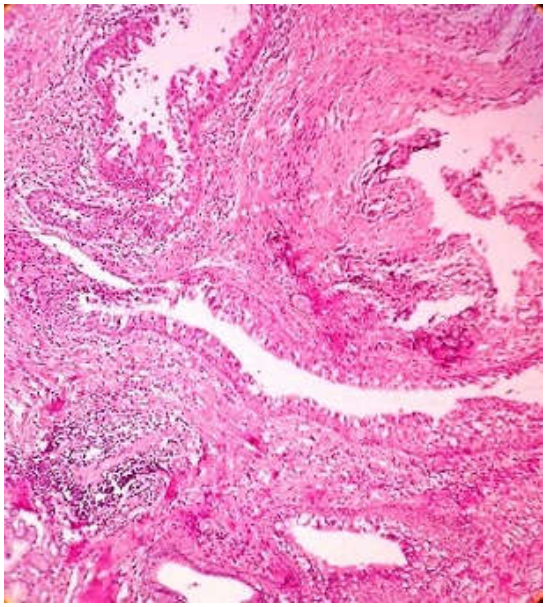


Fig.3. Surgical Exposure by Enucleation

Fig.4. Removal of Lesion in Toto

Fig.5. Closure of the Surgical Wound

Fig.6 Post-Operative Extraoral View



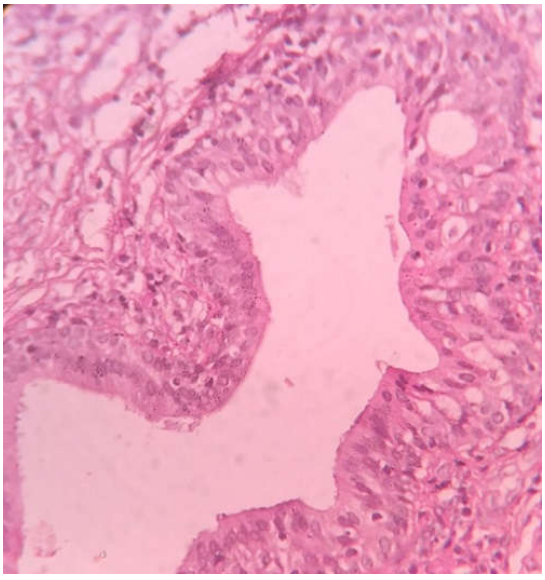


Fig.7 & 8. Microphotograph Showing Many Cystic Spaces Lined By Mucous and Epidermoid Cells. Connective Tissue Is Fibrous With Mild Inflammatory Cells.(Hematoxyline & Eosin X 100, X 400)

DISCUSSION

Mucoepidermoid carcinoma (MEC) was first described by Volkman in 1895, which was further elaborated upon by Stewart in 1945 as mucoepidermoid tumor. The credit of naming the tumor as MEC goes to Foot and Frazell (1953). The MEC can be associated with major salivary glands, minor salivary glands, and can also occur as an intraosseous tumor frequently called as central MEC (Ranganath *et al*., 2011). The common age group for the occurrence of MEC in males and females is around 43 years with an overall female predilection (76–80%). The gender difference is extremely pronounced in patients with lesions of the tongue and retromolar area. The 46% of MEC's occurring intraorally in the minor salivary glands arise in a variety of location including ectopic salivary gland tissue. Most of the cases are frequently seen to be associated with palate, cheek, mandible, lip, and tongue. Lesser number of MECs are associated with the labial mucosa, retromolar area, oropharynx, and ectopic salivary gland (Ranganath *et al*., 2011). The etiology of carcinoma of the salivary glands includes alcoholism, smoking, exposure to radiation, viral infections, diet and nutritional insufficiencies. In our case the probable cause appears to be chronic irritation of the mucosa which directs towards an uncommon etiological occurrence for MEC. In MEC, the recurrence rate is approximately 25% and 10% in low grade lesions. Metastasis and survival rate are related to the histological grade and stage. The treatment for MEC in minor salivary glands is primarily surgical. A wide excision with removal of underlying bone if involved is recommended (Ranganath *et al*., 2011). The case presented here is a rare and unusual case, presenting as bilateral swellings in an atypical gender and age group. The clinical characteristics indicated of the pathology to be mucoceles. The patient also gave a history of lip biting which indicated that the lesion was a result of chronic irritation. Our patient presented with swellings seen in labial mucosa bilaterally since 1 year which had a localized fluctuant nodule like appearance. The clinical and radiographic differential diagnosis of a labial mucosal mass includes mucocele, which is frequently seen in fluctuant reactive lesion of salivary glands, with transparent blue swelling including mucin with a history of chronic lip biting. Malignancy seen in salivary gland tumors is 50% in children and 15-25% in adults. As a typical intraoral presentation this malignancy has a painless and persistent enlargement, which lasts for about a year. Intraoral lesions with paresthesia, pain, and difficulty with swallowing are noted frequently when major salivary glands and tongue are involved (Kulkarni *et al*., 2014). High-grade lesions may be quite firm and accompanied with ulceration, resorption of bone, and numbness of adjacent teeth, these clinical findings were not presented in our case. High-grade MEC epithelial cells are predominantly squamous, whereas in low-grade, mucous cells predominate. There was no significant radiographic finding found in our case as it was completely in the soft tissue of the skin. The presentation of our case reported was more toward benign nature.

Low grade MEC's of the oral minor salivary glands generally have a good prognosis. Moreover the central MEC and the one arising from minor salivary glands are usually of low-grade variety. Low-grade tumors are soft and compressible. This

finding correlated with our histopathological report which showed mucous cells predominantly. The tumor should be excised to obtain adequate tumor-free margins. However, if there is any evidence of bony involvement, removal of a portion of the jaw is necessary (Kulkarni *et al*., 2014). In our case since there was no bony involvement, only wide local excision was advocated. Chemotherapy was not used as an adjuvant therapy in our patient and does not currently have a role in the standard treatment of MEC patients (Kulkarni *et al*., 2014). Overall survival rate has been linked to histocytologic grade with 95-100% in low-grade and 25-43% in high-grade tumors.

Also it should be considered that treatments like micro marsupialization, cryosurgery, and laser therapy are contraindicated in management of an intraoral submucosal mass or nodules, as they may result in local spread of the tumor, and more aggressive surgery may be needed. Based on low recurrence and mortality rates in surgical treatment of low-grade MEC, the treatment of our patient is adequate. Follow-up of the patient was done for 1 year and the patient has not reported with any sign of recurrence or any other related complaints. Survivors of the adolescent cancer must be followed closely throughout their lifetime for the risk of developing a secondary malignancy following the treatment of adolescent cancer.

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Patient permission: Obtained

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