Case Report:

A Rare Case Of Mucoepidermoid Carcinoma Presenting As A Rare Longstanding Mucocele

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Abstract
Mucoepidermoid carcinoma is the most common malignant neoplasm found among major and minor salivary glands. Intraorally it shows more predilection for palate. Also other common sites include buccal mucosa, tongue and retromolar area. Tumour of low-grade malignancy appear as slow growing, painless mass that seldom exceeds 5 cm in diameter. Here we report a rare case of mucoepidermoid carcinoma which was initially diagnosed as mucocele. The lesion was asymptomatic for 13 years and was then histopathologically confirmed to be a low grade mucoepidermoid carcinoma during a routine dental check-up.
Keywords:-Mucoepidermoid carcinoma, Mucocele, Buccal mucosa, Low grade, Tumour

الملخص
سرطان الغشاء المخاطي هو الورم الخبيث الأكثر شيوعًا بين الغدد اللعابية الكبرى والثانوية. داخليا فإنه يظهر المزيد من الميل للحنك كما تشمل المواقع الأخرى الشائعة الغشاء المخاطي الشوكي واللسان ومنطقة الشبكية. يظهر هذا السرطان كورم خبيث منخفض الدرجة وبطيء النمو وغير مؤلم لا يتجاوز قطره ٥ سم. هذه حالة نادرة من سرطان الغشاء المخاطي الذي تم تشخيصه في البداية على أنه تورم بالغشاء المخاطي. كانت هذه الحالة بدون أعراض لمدة ١٣ عامًا، ثم تم تأكيدها من الناحية المرضية على أنها سرطان غشاء مخاطي منخفض الدرجة أثناء فحص الأسنان الروتيني.

Introduction

Mucoepidermoid carcinoma which is a malignant epithelial tumour was first studied by Stewart, Foote and Becker in the year 1945. As the name suggests, the tumour consists of both mucus secreting cells and epidermoid type cells in different proportions. Mucus cells are seen in clusters or they can occur singly. Mucus cells have a pale, at the same time a foamy cytoplasm. They have a distinct cell boundary and also a small peripherally placed compressed nuclei. Epidermoid cells with their eosinophilic cytoplasm may not be common but can sometimes show keratin

pearl formation. Often another group of cells which doesn't belong to mucous or epidermoid type can also be seen. They are intermediate cells which either small basal cells with scanty basophilic cytoplasm or oval cells with more abundant pale eosinophilic cytoplasm. Columnar and clear cells are also present

Parotid gland is usually most commonly affected [1]. This carcinoma of salivary gland accounts for about 5% of all salivary gland tumours. It occurs in individuals from second to seventh decade. While it is rarely seen in the first decade of life [2]. In children, mucoepidermoid carcinoma is the most common malignant salivary gland neoplasm. Usually, they are of three grades; low, medium and high. Low grade tumours have a limited potential to metastasize to regional lymph nodes. Due to their tendency to develop cystic areas, mucoepidermoid carcinoma shows close resemblance to mucocele, especially those of the retromolar region. We report a rare case of mucoepidermoid carcinoma which was asymptomatic for about 13 years and was initially thought to be a mucocele.

Case report

A 58-year-old male patient reported to the Department of Oral Medicine and Radiology, Sree Anjaneya Institute of Dental Sciences, Calicut for routine dental check-up. A swelling was noticed on the right buccal mucosa since 13 years. It was initially small in size and gradually increased to its current size with periods of remission and exacerbations. Swelling was completely asymptomatic during these years. The past medical history and dental history were not significant for our case.

On intraoral examination, a solitary, sessile, round, swelling was seen on the right buccal mucosa adjacent to lower vestibule in relation to 45 and 46 which measures about 1.5cm in diameter (Figure 1) with well-defined margins and normal colour of the mucosa. On palpation all the inspectory findings were confirmed. The swelling was non tender, soft, fluctuant with a brownish discharge. Adjacent teeth showed no mobility or displacement. Involved teeth responded to electric pulp testing. Panoramic radiograph showed no bone involvement. Based on the history and clinical findings we came to a provisional diagnosis of mucocele. Excisional biopsy was done, and the specimen was sent for the histopathological evaluation of the lesion.



Figure (1) solitary, well defined, round swelling in the right buccal mucosa adjacent to the vestibule in relation to 45 and 46

Histopathology revealed the presence all the three cellular types in the connective tissue (Figure 2). Mucous cells had an abundant, pale and a foamy cytoplasm. Epidermoid cells demonstrated a polygonal shape and they had intercellular bridges. Few of them showed keratinization. Basaloid cells referred to as intermediate cells were also present. Epidermoid cells together with intermediate and mucous cells were lining the cystic spaces. Overlying normal epithelium showed cystic spaces. Clusters of clear cells were also present. Clear cells and intermediate cells were sheet pattern.

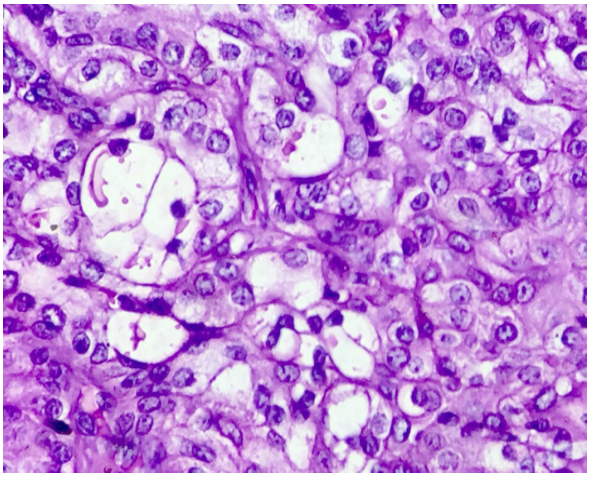


Figure (2) Microscopic view 10X - Connective tissue showing predominantly mucus cells with abundant, pale, foamy cytoplasm. Cystic areas can also be seen

Discussion

Among the salivary gland neoplasms, 10-15% originates from minor salivary glands [3]. Usually patients complains of a longstanding (more than 12 months generally) painless swelling in the mouth. Intraoral mucoepidermoid carcinoma affecting the minor salivary gland can even occur in the ectopic salivary gland tissues. Palate, cheek, mandible, lip and tongue are the common sites whereas frequency of mucoepidermoid carcinoma is less in retromolar area, oropharynx, and ectopic salivary gland.

This is the first case of mucoepidermoid carcinoma reported in the literature with a longstanding history of 13 years. Mucoepidermoid carcinoma can be completely asymptomatic during the course of progression. It may present itself as a bluish, red – purple, fluctuant, smooth surfaced mass closely resembling a mucocele. The present case is similar to this. The middle aged male patient showed a prolonged, at the same time slow progression of the tumour. Localised fluctuant swelling was present for nearly 13 years which at times showed episodes of regression and then reappearance. The patient reported no symptoms associated with the lesion. Major salivary gland involvement can result in pain, paraesthesia, and other difficulties with the lesion. When low grade tumours are soft and also compressible, high grade tumours show ulceration, bone resorption and adjacent tooth numbness. All these clinical features are not there in our case. Features of high grade tumours are absent in our case. Hence it was necessary to rule out the possibilities for a low grade tumour also. Unfortunately, histopathological reports were however favouring the chances of a low grade mucoepidermoid carcinoma rather than a mucocele.

Pluripotent reserve cells of excretory ducts can differentiate into squamous, mucous, or columnar cells and can give rise to mucoepidermoid carcinoma [4,5]. Following the histopathological studies, based on the proportion of all the three cell types and by the extent of atypia presented by cells, MEC can be categorized into three grades. Low grade, intermediate grade and high-grade tumours. As the name suggests low grade tumours present minimal cellular atypia at the same time high grade tumours have considerable amount of cellular atypia. Mucous cells predominate in low grade tumour whereas high grade tumours show islands or sheets of intermediate and epidermoid cells. Intermediate grade tumour shows more of intermediate cells and degree of cellular atypia is more than low grade but less than a high grade tumour.

Wide local excision ensures oncologic safety in case of low grade and intermediate grade tumours [6,7,8]. Treatment plan is made depending upon the location of tumour, aggressiveness of the tumour mass. Treatment shall however result in tumour free margins. If tumour has progressed to that extent where bone is involved, the affected bone should also be removed. As low grade tumour shows less recurrence, we planned surgical excision of tumour mass in our case. High grade mucoepidermoid carcinoma require more invasive surgical treatment or even chemotherapy or radiotherapy following that. Low grade and intermediate grade mucoepidermoid carcinoma show very low recurrence rate that is less than 10%. At the same time the survival rate is about 90%. Radical neck dissection is indicated in case of metastasis. Low and intermediate grade tumours rarely show metastasis. Grade of the tumour determines the prognosis [7,8]. We followed up our patient for one year. There were no signs of recurrence or any other complaints.

Conclusion

One should be kept in mind that mucoepidermoid carcinoma should be considered as a differential diagnosis for longstanding cases of mucocele. This is the first case of mucoepidermoid carcinoma in the literature reported with a longstanding history of 13 years. Proper clinical and histopathological evaluation is mandatory for the proper diagnosis and treatment planning.

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