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Case Report

Mucoepidermoid Carcinoma: a rare case report

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Abstract: Salivary gland tumors are generally considered to be one of the most difficult areas in the diagnosis of oral lesions. mucoepidermoid carcinomas (MECs) are the second most common malignant salivary gland neoplasm. Here in we report a case of a low-grade MEC of the sublingual salivary gland in a 35-year-old woman. Wide excision of the tumour along with continuous marginal mandibulectomy was performed. The treatment outcome was excellent .There has been no recurrence till date.
Keywords: salivary gland, MECs, oral lesions.

INTRODUCTION

Mucoepidermoid carcinoma is commonly found in salivary glands and accounts for 5–10% of all salivary gland tumours [1-3]. Mucoepidermoid carcinoma is a kind of malignant tumour of epithelial origin consisting of different proportions of, epidermal, mucous, intermediate, and clear cells and often shows cystic growth. Mucoepidermoid carcinoma is generally differentiated as low, intermediate, or high grade on the basis of its histology, which includes the presence or absence of cystic spaces, cellular differentiation, area occupied by mucous cells, growth differentiation, invasion, and cytologic atypical features. In the year 1945, Stewart et al described its mucous-secreting and epidermal cellular elements thus identifying it as a definite pathologic entity. Eversole reviewed 815 cases and found that of the major salivary gland tumours, 89.6% are associated with parotid gland, 8.4% with submandibular and 0.4%with sublingual gland [1]. The palate was the most common site for minor salivary gland involvement that accounts for 41.1% of intraoral lesions [1].

CASE PRESENTATION

A 61-year-old Indian male reported to the Department of Oral Medicine and Radiology, with the chief complaint of a swelling in the floor of the mouth since 2 weeks. He had noticed a small, painless swelling on the floor of the mouth, which had grown to the present size. He had difficulty in mastication, swallowing and talking. No relevant medical history

Intraoral inspection revealed a solitary, well-defined, round swelling, on the floor of the mouth was ulcerated and bleeding was present. It was firm in consistency, with well-defined borders, movable and tender (figure 1) the radiological investigations like OPG are non contributory. A clinical diagnosis of a tumour of minor salivary gland neoplasm was arrived.

Histopathology

Laboratory investigations revealed a normal hemogram. Intraoral defect on the floor of the mouth was reconstructed using a left lateral tongue flap. Microscopic picture of the tumour showed abundant cystic spaces surrounded by mucous cells and few epidermoid cells. Based on the histopathological findings (according to WHO staging of salivary gland tumours, 2005), a final diagnosis of a ‘low-grade mucoepidermoid carcinoma of the sublingual salivary gland’ was suggested (Figures 2 & 3).

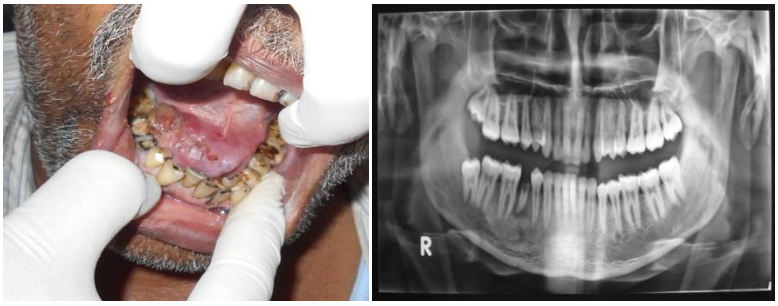

Fig-1: Intraoral figure showing a well-defined swelling on the floor of the mouth along with OPG of the patient

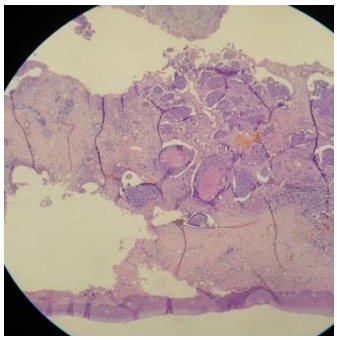

Fig-2: Histopathological picture- abundant cystic spaces surrounded by mucous cells and few epidermoid cells seen.

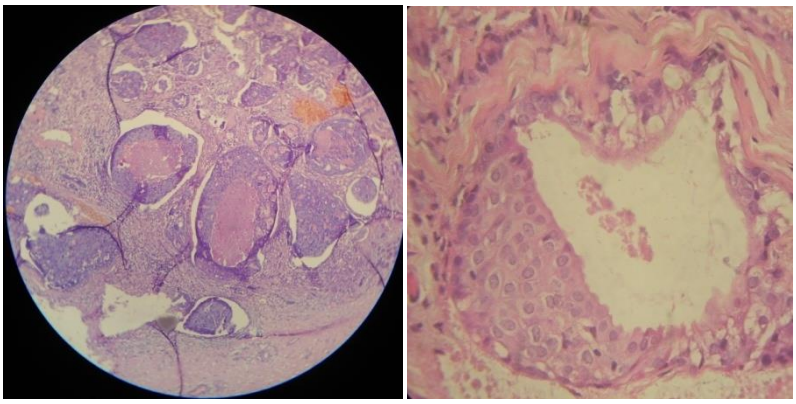

Fig-3: Histopathological picture – 40× magnification of histopathological specimen.

DISCUSSION

Among malignant tumours in salivary glands, 15–32% is of parotid, 41–45% is from submandibular gland, 70–90% from tumours of sub lingual salivary glands and 50% in minor salivary glands. the carcinomas in sublingual glands accounts for about 1% of total major salivary gland carcinomas, and around 0.5–1% of all epithelial salivary tumours origin [2,4]. 80 to 90% of sublingual salivary gland tumours are frankly malignant, and MEC was found to be the most common malignant salivary gland tumour (ie, it accounts for 30% of total malignancies in salivary glands, 10% of total carcinomas in salivary glands and 5% of head and neck salivary gland cancers) [1, 4] about 50 to 60% of MECs derived from the major salivary glands of which only about 2–4% from the sublingual salivary glands, (the total 80% evolves from parotid and around 8–13% from the submandibular). It is also found to occur in minor salivary glands (most often in the palate) and in the seromucous sites of trachea, sinonasal tract and larynx [1].

Smith et al. reviewed article on the mixed tumours of the salivary glands, concludes that in a research by Dr BR Shore in St. Luke's hospital, only one sublingual variety of salivary gland tumour was seen during the past 40 years.[4] Goode and colleagues in their research of 234 cases of MEC in the major salivary glands found around 3% in the sublingual gland only [5]. Ma'aita and colleagues studied all the variety and clinical results of 21 salivary gland tumours in Jordan and reported only 1 case (0.4%) of sublingual type of salivary gland tumour.[6]Poomsawat *et al* in a retrospective study of 60 cases of salivary gland tumours in a Thai population concludes that 10% are in major salivary gland tumours. Of these six cases, two cases had its evolution form sublingual glands.[7]MECs have no genetic origin,nor they have any relation with sialoliths, trauma, any abuse of tobacco or alcohol.

Primary tumours of the sublingual salivary gland are uncommon and usually seen as a slowly increasing mass situated beneath the tongue. The benign mass may be present as a painless lesion for many years whereas a malignant tumour may be present from a few months to a year and may present with pain when perineural, cartilage or bone invasion is questioned. Malignant tumours are usually found around the 3rd to the 5th decade of life, with a slight predeliction for female [1].

Goode *et al* found that 80% of patients with sublingual gland tumours were symptomatic [5].

Differential diagnosis includes:

- The differential diagnosis includes neoplasm arising from submandibular/sublingual gland, mucocele sialolithiasis and Kuttner's tumour.
- Mucocele, low-grade MEC and mucous cell adenocarcinoma both produce mucus and thus simulate each other.
- Sialolithiasis is a very common pathology that affects the submandibular gland/duct.
- Kuttner's tumour is type of reactive disorder in the submandibular gland which clinically is seen as a firm painful indurated enlarged gland. Its nearness to sublingual gland jutifies its inclusion.
- Mucinous adenocarcinoma is an entity frequently found in palate and sublingual gland and hence needs to be excluded.

Plane radiographs, sialography and nuclear scans are of little diagnostic importance and are rarely indicated for evaluation of the sublingual gland masses. Sonographically, MECs, which are smaller than 2 cm, have a uniform structure with smooth borders and may wrongly, be diagnosed as benign. CT and MRI provide a clear visualisation of the salivary glands. CT scans are especially useful in identifying the location and density of the lesion. They help to demarcate cystic from solid lesion and in assessing the relation with nearby structures including the bones and soft tissues [1].

Ideally on histological examination MEC's contain three cellular elements in different proportion: squamous cells, mucus-secreting cells and the intermediate cells. The appearance changes with the type of the tumour. The WHO classification (2005) proposed that the tumours of the salivary gland should be classified as low, intermediate and high-grade salivary gland tumors [2]. The low-grade variety has an increased ratio of mucous cells and is less aggressive lesions. While high-grade form is considered to be more malignant tumour and carries a poorer prognosis, [4,8] low-grade tumours usually have well-circumscribed ovoid lesions of 2–4 cm in diameter which contain solid grayish white or greyish pink regions which are a mixture of mucus-filled macrocysts, very few solid nests of intermediated and epidermoid cells and no cellular pleomorphism [9].

Treatment

Surgery is the mainstay of treatment. Brookstone and Huvos concludes in their research that 40% reappearance or recurrence post conservative surgical techniques such as curettage, enucleation, marsupialization and marginal resection with or without adjuvant therapy .in this case enucleation was performed. Adjuvant radiotherapy is recommended for high-grade tumours [10]. Patient is disease free and currently under regular follow-up.

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