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Case report

Mucoepidermoid Carcinoma of Palate Treated with Temporalis Muscle Flap Reconstruction

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Abstract

Mucoepidermoid carcinoma is a malignant salivary gland tumor that develops from the multipotent cells, structural units, of the excretory canal. Generally observed in parotid gland, followed by minor salivary glands. Histologically, it can be categorized into low, intermediate, and high grades and is made up of mucus, squamous, and intermediate cell types. We present a case of Mucoepidermoid carcinoma in in the left posterior area of the palate in an 18-year-old male reported to department of Maxillofacial, The National Cancer Institute, Misrata, Libya. The lesion treated by wide local excision with adjacent free margins and left subtotal maxillectomy followed by reconstruction with temporalis myofascial flap. Prophylactic neck dissection at level I-III was done. The objective of this case report is to present the clinical manifestation, diagnosis, and proposed treatment plan of mucoepidermoid carcinoma of the palate. This case report emphasizes the significance of an accurate diagnosis and treatment strategy when dealing with malignant tumors because failure to do so can result in morbidity and fatality. The severity and aggressiveness of the lesion determine how mucoepidermoid carcinoma should be treated.

Keywords: *Diagnosis, Salivary Gland, Mucoepidermoid Carcinoma, Temporalis Flap, Subtotal Maxillectomy.*

Introduction

Mucoepidermoid carcinoma is an epithelial tumor typically seen in the salivary glands. This neoplasm makes up between 2.8% and 15% of all malignant salivary gland tumors, making it the most frequent one (1). Only 2% to 4.3% of all mucoepidermoid carcinomas described in literature are abnormal salivary gland neoplasms that develop within the jaws as primary intraosseous (central) bone lesions (2). A rare but well-known type that involves the jaw bones is central mucoepidermoid carcinoma.

The majority of primary central mucoepidermoid carcinomas develop in lower jaw, while maxilla lesions are uncommon (3). There has been a lot of discussion on its etiology, radiological, and histological aspects (4). Rarely, it could develop intraosseously from the epithelial lining of the odontogenic cyst or epithelial remnants of salivary glands grew ectopically (5). We describe a rare instance of a primary intraosseous mucoepidermoid carcinoma in the posterior palatine region in a male patient who was 18 years old. At the National Cancer Institute, Misrata, Libya the lesion treated by wide local excision with adjacent free margins and left subtotal maxillectomy followed by reconstruction with temporalis myofascial flap. Prophylactic neck dissection at level I-III was done.

Case Report

An 18-year-old male patient presented to maxillofacial unit at the national cancer institute of Misrata complaining of ulcerative lesion at the inner aspect of his upper jaw on the left side posteriorly that had been present for one year. Patient reported that the lesion started as minor swelling which was asymptomatic and does not interfere with eating, but it progressively grew larger and became uncomfortable. Fifteen days before his presentation to our

clinic, incisional biopsy was taken at a private medical center and histopathology result confirmed presence of mucoepidermoid carcinoma. No history of previous illness, loss of appetite or weight loss was mentioned. Patient denied smoking or alcohol consumption.
Upon examination, an ulcerative lesion measuring about 3 by 2 cm was found in the left palatal area, directly in face to upper premolar and molar teeth. The overlying mucosa was ulcerated and infected (Figure 1). All teeth on this side were apparently free of caries. Also, head and neck lymph nodes were neither tender nor palpable.

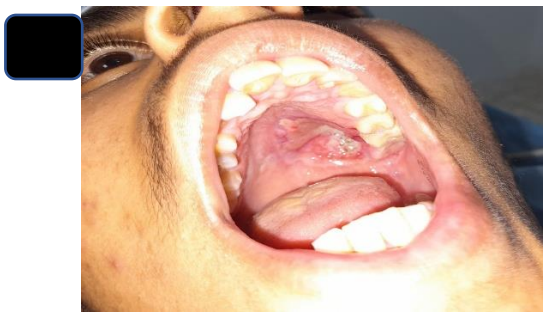

Figure 1: Clinical presentation of the lesion: endophytic ulcerative lesion located in the left hard palate and extending to the soft palate, crossing the midline.

A well-defined unilocular radiolucency with sclerotic borders was visible on an orthopantomographic view radiograph in the premolar molar region (Figure 2).

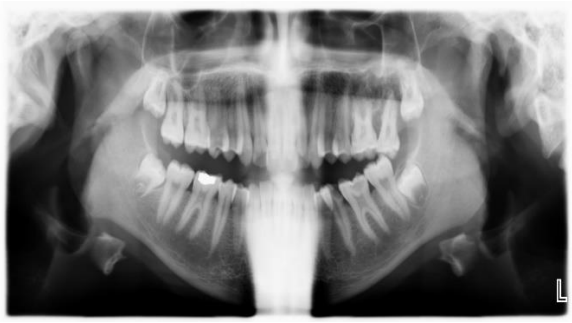

Figure 2. Orthopantomography view showing a well-defined unilocular radiolucency with sclerotic borders at the premolar molar region.

A contrast-enhanced computed tomography (CE-CT) scan of the skull base, neck, chest, abdomen and pelvis revealed left hard palate/left maxillary sinus floor are seat of bone destructive mass lesion 3.8x2.7cm, with no associated sizeable lymph nodes or distant organ metastasis (Figure 3).

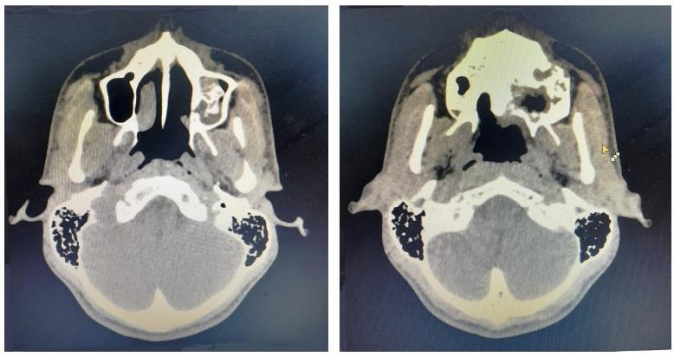

Figure 3. Maxillary axial CT scan at two different levels revealing an expansile destructive mass in the left side.

Left subtotal maxillectomy with temporal muscle flap reconstruction along with neck dissection was part of a planned treatment for the lesion. Under general anesthesia, in the strictest aseptic and sterile measures, the patient was readied for surgery.
Weber Ferguson approach and left submandibular incision were performed. The palatal lesion was outlined and mucosa was excised with safe margin of (1cm). Palatal bone and left lateral wall of maxilla was resected using surgical saw. With the aid of a pterygoid chisel the pterygomaxillary dysjunction was done and the lesion was detached from the rest of maxilla (Figure 4).

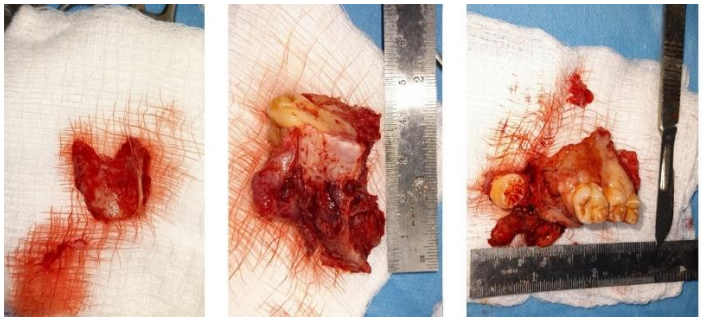

Figure 4: Resected lesion specimens.

On the left temporal area, a modified Alkayat Bramley incision was made. Subgaleal flap was raised till the inferior border of the zygomatic arch was reached. At the superior temporal line, the temporalis muscle was incised and lifted from the pericranium (Figure 5 and Figure 6). Temporalis muscle was tunneled under the zygoma (Figure 7) to reconstruct the left class Ib defect of the palate and maxillary region according to Okays classification (Figure 8).

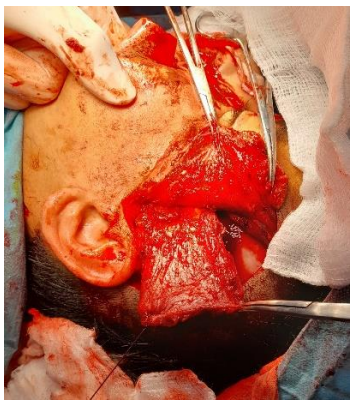

Figure 5: Temporalis myofascial flap incised and separated from the overlying skin.

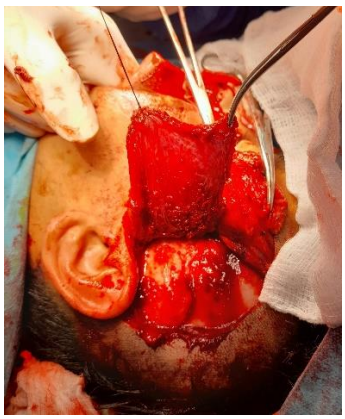

Figure 6: Temporalis myofascial flap elevated.

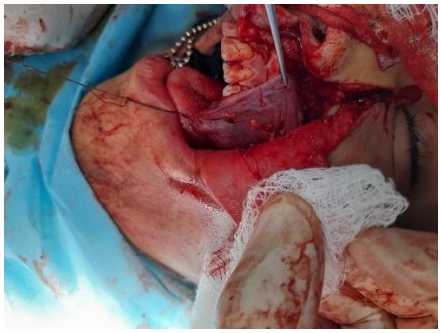

Figure 7: Tunnelling of the flap below the zygomatic arch intraorally.

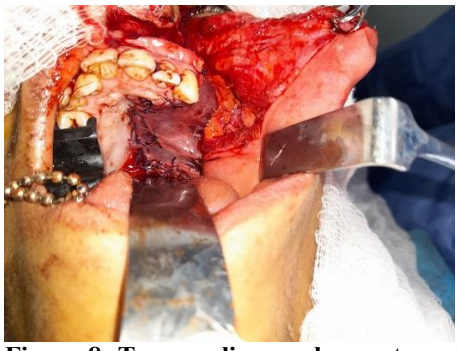

Figure 8: Temporalis muscle was tunneled under the zygoma to and sutured in place to cover the left Class II (d) defect of the maxilla.

Left level I -III neck dissection was done. The supraomohyoid neck dissection (a selective cervical node dissection) was performed in which the contents of the submental and submandibular triangles (lymph node level I), the jugulodigastric and jugulo-omohyoid lymph node groups, and the lymph node-bearing tissues located anterior to the cutaneous branches of the cervical plexus and above the omohyoid muscle (lymph node levels II and III) were removed (Figure 9).

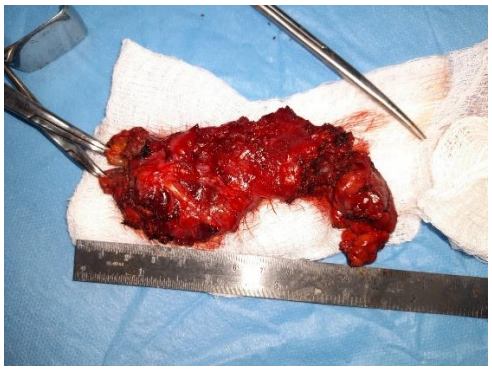

Figure 9: Resected lymph node specimen of neck dissection.

Hemostasis is attained. In the left temporal and left neck areas, a closed wound suction drain was inserted in place. Using a 2-0 Vicryl round body, reverse cutting silk, the intraoral and extraoral closure was completed. Throughout the entire surgery, there were no complications. Resected tissues were sent for histopathological analysis. Results of histopathology examination revealed that tumor is composed of sheets of squamous cells along with cystic spaces lined by mucinous epithelium. Tumor islands are separated by fibrocollagenous tissues. No lymphovascular or perineural invasion was seen. In the dissected lymph nodes, metastatic disease was not seen. The diagnosis of a low-grade mucoepidermoid carcinoma was confirmed.

Follow-up
In order to check for any recurrence, the patient was under periodic review at 1 week, 2 weeks, and 15 days for 3 months and later every month for 1 year during which he remained asymptomatic. Healing was unremarkable after 2-year of follow up (Figure 10). CT scan for the neck with intravenous contrast showed stationary interval with no fresh lesion seen. The patient has been receiving monthly follow-up for two years without any signs of a recurrence. Patient received further adjuvant therapy according to mucoepidermoid carcinoma, grade 2, hard palate.

Outcome
With a follow-up period of two years, the patient is performing well and was lesion free, prognosis was excellent. Mouth opening, and the graft recipient and the donor sites were symptom free with normal healing (figures 21 and 22).

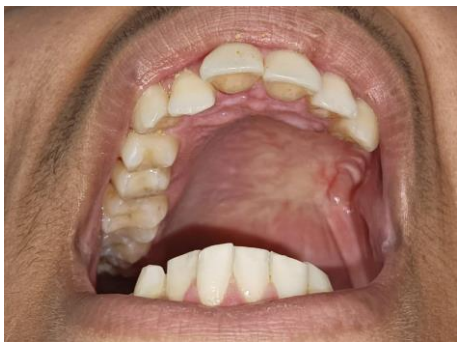

Figure 10: Intraoral view showing complete healing of oral mucosa at surgical site after two-year of follow up.

Discussion
Primary intraosseous mucoepidermoid carcinoma is an infrequent tumor (6). It has been proposed to be encompassed in the primary intraosseous carcinomas of the jaws as type 4 (7). The fourth and fifth decades are the most prevalent for Primary central mucoepidermoid carcinomas to arise. However, cases have been documented to develop also in the first to seventh decade (8). In the present case, our patient is comparatively young, he was 18 years old. Although this type of tumors is slightly more prevalent in females and occurs in the posterior mandible in adults (8), this patient was a male, and the site was the palate.
Mucoepidermoid carcinoma typically manifests as an asymptomatic bump. The symptoms of pain, paresthesia, numbness, and tooth movement are frequently late discoveries (9).
Mucoepidermoid carcinomas are classified as malignant using the criteria of Seifert and Sobin (10) and Auclair et al. (11) Table 1.

Grade	Criteria
Low grade:	Highly differentiated neoplasia with a predominance of macro and microcysts. Presence of intermediate and mucin-producing cells,
Intermediate grade:	Predominance of intermediate cells and a few cysts. Presence of mucin-producing cells and islands of epidermoid cells,
High grade:	Poorly differentiated neoplasia with predominance of intermediate and epidermoid cells in solid blocks. Mucin producing cells are present.

Our patient found to has a low-grade mucoepidermoid carcinoma. Histopathologically, low-grade mucoepidermoid carcinomas are made up primarily of cuboidal to columnar mucus-secreting cells that are clustered around microcystic structures, interspersed with a small number of epidermoid cells. Small cysts coalescing into larger cystic areas is indicative of low-grade cancer.
Numerous potential origins have been taken into account when analyzing the pathophysiology of the central Mucoepidermoid carcinoma (12, 13), including: (1) Neoplastic transfor-

mation of the mucus-secreting cells from the epithelial lining of the dentigerous cyst associated with impacted third molars, (2) embryonic remnants of the submandibular and sublingual glands trapped within the mandible during development, (3) neoplastic transformation and invasion from the lining of the maxillary sinus, (4) neoplastic transformation of the mucus-secreting cells from the epithelial lining of the dentige

Nevertheless, as central mucoepidermoid carcinoma of the maxilla is a rare lesion, no much data is available about its pathophysiology. The following are some potential sources of the central mucoepidermoid carcinoma in this patient: (1) invasion and neoplastic transformation from the maxillary sinus lining, and (2) entrapped minor salivary gland neoplastic transformation.

A radiographic examination is crucial for making a differential diagnosis, determining whether the condition qualifies as a central jaw lesion(14), as well as its magnitude and any involvement or encroachment on nearby important tissues. A three-grade classification for intraosseous mucoepidermoid carcinoma was proposed as the following(9): Grade 1, without expansion and rupture of the cortical plate; Grade 2, with expansion but without rupture of the cortical plate; and Grade 3, with rupture of the cortical plates or the presence of regional metastasis. Our patient matched Grade 3 with a cortical plate rupture.

For patients with central intraosseous mucoepidermoid carcinoma, surgery including curettage, enucleation, marsupialization, and extensive local excision, is the main form of treatment (9). Soft tissue palatal excision with peripheral margins of 1 cm and anatomical barrier margins is necessary for low-grade mucoepidermoid carcinomas that develop in the palate (15) The palatal bone need not be removed unless radiography, scans, or direct examination reveal malignant extension into it. However, mucoepidermoid carcinoma should typically be treated with extensive local excision, en bloc resection, hemimandibulectomy (9), or hemimaxillectomy, even when they are low-grade tumors.

In situations when metastasis to the cervical nodes is suspected, neck dissection is typically a component of the treatment (16). prophylactic neck dissection is not recommended. Cases of high-grade mucoepidermoid carcinoma are advised to receive radiotherapy (16, 17). Postoperative radiation is not recommended for low-grade tumors unless vital structures are involved. Although central mucoepidermoid carcinoma cases should be followed up for a longer period of time up to 10 years due to the chance of late recurrence or regional metastasis, these tumors typically have a good overall prognosis (16). However, if the expansion affects crucial areas like the base of the brain, death could happen. The literature contains very little information regarding the recurrence rate of central mucoepidermoid carcinoma (18). When oral mucoepidermoid carcinoma of low grade is treated in this way, the 5-year survival rate is approximately 95% (15).

It is only for mucoepidermoid carcinomas that histologic grading of the tumor and clinical attitude traits are related and partially predictive of the observable response of the patient.

The histologic grade is consequently used to guide treatment options. Low-grade mucoepidermoid carcinomas have an infiltrative growth pattern and a very slow rate of growth, similar to a pleomorphic adenoma. Rarely, and only at the end of their progression, can they metastasis. High grade mucoepidermoid carcinomas, on the other hand, develop and spread quickly by infiltration like poorly differentiated squamous cell carcinomas (19). The temporalis muscle flap is substantial. Despite the development of microvascular flaps, it nevertheless offers a very good healing and functional outcome for repairing imperfections of palate. Given that the flap is extremely strongly vascularized, necrosis rates are minimal. It can cover significant and even bilateral palate abnormalities and delivers good functional outcomes for speech and swallowing. It can be cut down utilizing a variety of little and large incisions. Small incisions are sufficient in situations where the only thing needed to seal a defect is a strip of muscle or fascia. In our case, the post-maxillectomy defect that resulted was a Brown's Class II d defect (20). Therefore, a large incision was made to harvest practically the whole temporalis muscle in order to properly cover a deficiency.

In addition, the surgical cut made to produce this flap can be used to get access to other tissue for repair, such as the temporo-parietal fascia, periosteum covering the temporal fossa's bone, and different bones forming the calvaria. This type of flap can be effective to cover unilateral deformities that are up to 6 cm long and bilateral deformities that are up to 4 cm long that cover the whole palate (pre-maxilla with hard and soft palate) (21).

Complications-free closure of palate deformities up to 6 × 5 cm in size, covering both the hard and soft palate has been reported previously (22). The temporalis muscle flap can be used for the following conditions, according to Brown and Shaw's recommendations: vertical maxillary defects, provided they do not include the orbits or nasal bones; horizontal

palatal defects that are less than or equal to one-half the width of the palate; or a combination of the two (20).

In conclusion, we described a low grade mucoepidermoid cancer case that affected the hard and soft palates and was treated by a left lateral maxillectomy, wide local excision, and reconstruction with a temporalis myofascial flap. The patient received adjuvant Chemo-radio therapy. During a follow up period of two years, the patient is performing well. Consequently, temporalis flap might be viewed as one of the several treatment choices for palate deformities.

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