Case Report

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Medium-sized buccal mucosa defect reconstruction with buccal advancement flap in mucoepidermoid carcinoma ex pleomorphic adenoma: a case report

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Introduction and importance: Mucoepidermoid carcinoma (MEC) ex pleomorphic adenoma is a rare type of salivary gland cancer. Surgical resection remains the standard therapy for this malignancy. After tumor removal, larger defects may require a local, regional, or free flap, while smaller ones can be closed primarily. Managing medium-sized defects can be challenging, especially on the buccal mucosa.

Presentation of case: A 47-year-old man had a buccal mucosa mass for 10 years, which gradually grew over a year and irritated his chewing. A 2.2 × 2 cm buccal mass was observed with telangiectatic and erythematous alterations in the surrounding mucosa. The preoperative tissue biopsy suggested salivary gland malignancy. The patient underwent surgical excision and a single-stage buccal advancement flap reconstruction, successfully closing the 4 cm defect. The final diagnosis was MEC ex pleomorphic adenoma. He reported mild discomfort during the first few months while opening his mouth. The patient had fully recovered after 6 months.

Clinical discussion: This is the first case of MEC arising in a pleomorphic adenoma of the buccal mucosa. For low-grade and small-sized tumors, a single modality is appropriate for treatment. Local flaps such as buccal fat pad or musculomucosal flap can repair medium-sized defects. However, the buccal advancement flap provides effective functional and esthetic benefits, optimal healing conditions, and reduces complications risk.

Conclusion: The buccal advancement flap is a valuable option for reconstructing medium-sized buccal defects up to 4 cm. The single-stage surgical procedure has been proven to yield minimal complications and provide a favorable outcome.

Keywords: buccal advancement flap, buccal mucosa reconstruction, case report, mucoepidermoid carcinoma, pleomorphic adenoma

Introduction

Squamous cell carcinoma commonly affects the buccal mucosa, while salivary gland origin is less frequent. In rare entities, mucoepidermoid carcinoma (MEC) occupies most of them[1]. MEC originating from pleomorphic adenoma is exceedingly rare[2]. Tumor grading is divided into high, intermediate, and low grades[2]. For small, low-grade salivary gland cancer, complete surgical resection is the safest oncological treatment[3]. It is crucial to reconstruct the affected area after tumor removal to ensure the continuity of hold function and other vital functions such as physical, microbiological, and immune barriers[4]. Nevertheless, it is best to plan the restoration techniques carefully when dealing with medium to large-sized buccal mucosa defects and if the affected area involves other oral cavity subsites.

In this case study, a patient with low-grade MEC ex pleomorphic adenoma of the left buccal mucosa underwent wide surgical excision followed by immediate reconstruction. The medium-sized defect, which affected the buccal mucosa and upper lip, was successfully reconstructed using a buccal advancement flap. The case report has been reported in line with the SCARE 2023 criteria[5].

HIGHLIGHTS

- Mucoepidermoid carcinoma (MEC) ex pleomorphic adenoma is a rare tumor that typically occurs in the parotid gland.
- MEC ex pleomorphic adenoma originating from the buccal mucosa is an extremely rare condition.
- The buccal advancement flap is a single-stage procedure demonstrating functional and esthetic benefits.
- Buccal advancement flap technique is an effective method for reconstructing medium-sized buccal defects up to 4 cm.

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Case presentation

A 47-year-old man who worked as an agriculturist walked into the ENT (ear, nose, and throat) clinic by himself. He complained of a slow progressive left buccal mucosa mass that he had been experiencing for the past 10 years. The mass had gradually increased in size within the last year, causing mild discomfort to the patient while chewing. However, he denied any difficulty in eating, contact bleeding, or pain related to the mass. No history of smoking, betel nut chewing, alcohol drinking, denture wearing, or family history of head and neck cancer. There was no evidence of the patient having received any treatment for the buccal mass from other doctors. Additionally, there was no history of previous hospitalization or surgery related to other health problems, no underlying diseases, no current medication usage, and no history of drug allergies. During the physical examination, the patient's facial contour appeared normal, and there was no trismus. A firm, broad-based mass with a small area of central ulceration was identified on the left upper half of the buccal mucosa, measuring 2.2×2 cm. Surrounding mucosa manifested telangiectatic and erythematous alteration (Fig. 1). The tumor freely moved through the cheek's skin and subcutaneous layers. The patient was advised to have a biopsy of his buccal mass during the same visit to exclude the potential malignancy due to history and physical findings. Healthy tissue adjacent to the ulceration was biopsied with cup forceps, avoiding necrotic areas. The pathological results described dysplastic epithelium with marked acute exudative inflammation and necrotic area. Some tumor cells contained intracytoplasmic mucin, suspicious for salivary gland carcinoma. Differential diagnoses included MEC and polymorphous adenocarcinoma. The initial diagnosis was suspicious of salivary gland carcinoma. A computer tomography scan was requested to evaluate the extension of the disease, including the neck, and no evidence of cervical lymphadenopathy was found.

Although a definitive diagnosis could not be reached, the data suggested malignancy due to rapid tumor growth, abnormal surrounding mucosa, and pathological reports. Surgical resection with a wide margin and immediate single-stage buccal advancement flap reconstruction was planned and discussed with the patient. In our provincial general hospital, the general ENT surgeon who handled the case had to consider an additional permanent surgical margin due to the unavailability of intraoperative frozen sections. The defect size was 4×3.5 cm, mainly involving the buccal mucosa and a small upper lip area (Fig. 1). The anterior-based buccal advancement flap was designed to cover the entire defect (Fig. 2). Stensen's duct opening was well preserved. A Penrose drain was inserted to prevent fluid accumulation beneath the flap. On the fifth postoperative day, a minor wound dehiscence was observed at the posterior aspect, and a re-suture under local anesthesia was performed to correct the problem. The final pathological results reported carcinoma ex pleomorphic adenoma, which carcinomatous component suggests low-grade MEC. The carcinoma size was 6 mm in the greatest dimension. All margins were negative. Absent of perineural and lymphovascular invasion. The definite diagnosis was low-grade MEC (T1N0MO) arising in the pleomorphic adenoma. The first week after discharge, the patient visited the ENT office and reported experiencing mild discomfort while opening his mouth; however, no limitation mouth opening was observed. After 6 months of monthly appointments, all discomfort had disappeared, and he could open his mouth, drink, and chew normally (Fig. 3). The patient was satisfied with the surgery's functional and cosmetic results and scheduled a regular follow-up to ensure continued success.

Discussion

This is a unique case of MEC that occurred in the pleomorphic adenoma of the buccal mucosa. Based on the literature review, it

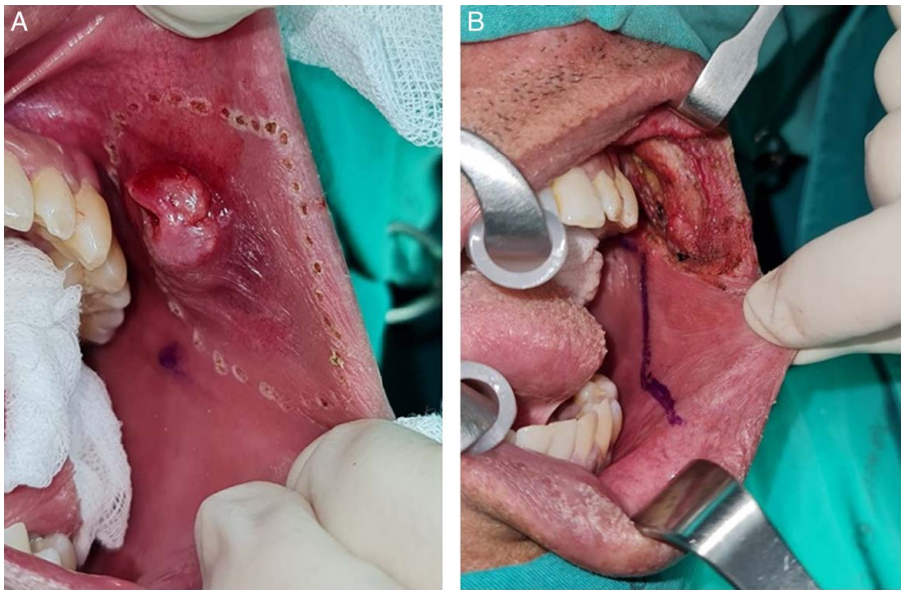

Figure 1. (A) Left buccal mucosal mass and its outline for surgical resection, Stensen's duct opening (marked in blue); (B) buccal mucosa defect with involved upper lip, the anterior-based buccal advancement flap outline (marked in blue).

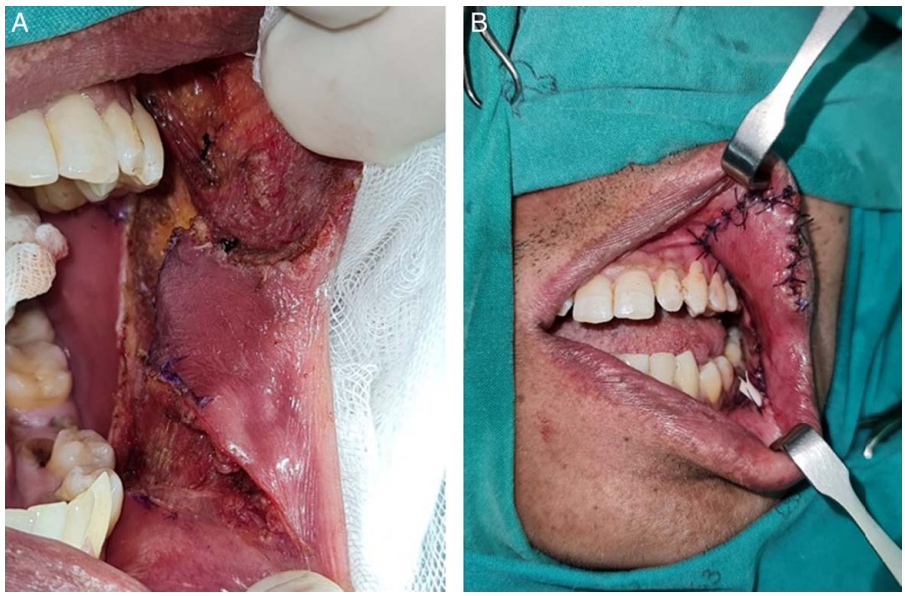

Figure 2. (A) The anterior-based buccal advancement flap harvested; (B) buccal advancement flap inset.

became evident that this discovery was the first of its kind in this specific location. Previously, a rare histology was commonly reported in the parotid gland[6–11], with only one case found in the submandibular gland[12]. The patient presented with a recurrent submandibular mass for 4 years after the excision of a pleomorphic adenoma[12]. A few cases were identified in the palate[2,13,14], including the hard palate[13], soft palate[14], and hard-soft palate junction[2]. In 2015, a rare tumor was discovered in a 26-year-old man's alveolar ridge and retromolar trigone[15]. Furthermore, there were three reported cases of lacrimal gland involvement[16–18]. A majority of the cases showed the presence of a high-grade tumor, indicating the need for a combination of treatment modalities[2,3,6–9]. However, the low-grade tumor was successfully treated by completely removing it as a single method[3,11], as shown in this case study. In this particular case, despite the absence of cancer risk factors, the rapid growth of the mass suggested that there was a possibility of malignant transformation, and it should be considered. In addition, according to the preoperative diagnosis of suspicious salivary gland carcinoma, surgical intervention was deemed the most optimal course of treatment. The primary objective of the surgical procedure was to remove the tumor, thereby ensuring clear surgical margins completely.

The precise reconstruction of the affected area following tumor removal is also important. This restorative measure facilitates the functional and esthetic rehabilitation of the oral cavity and minimizes the risk of potential complications. In cases where the buccal mucosa needs reconstruction, various factors must be considered. These factors include the size and placement of the defect, the extent of tissue loss, the underlying cause of the tumor, any additional medical conditions the patient may have, as well as the patient's attitude toward the procedure. Primary closure may

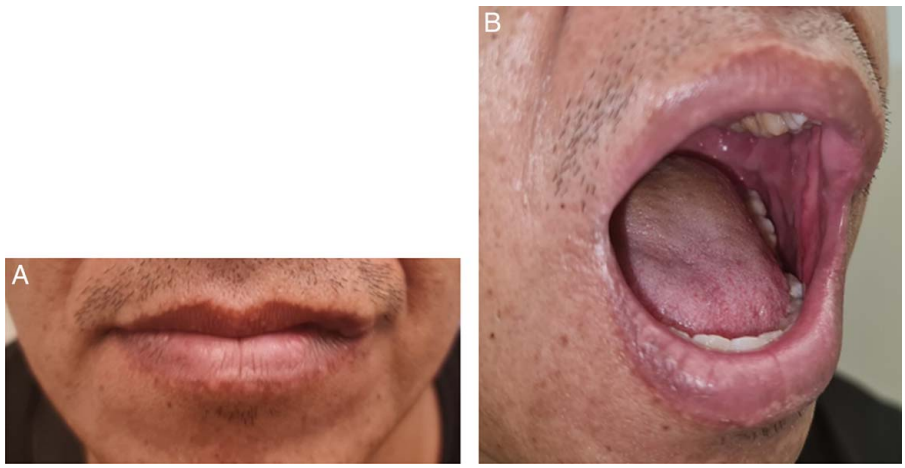

Figure 3. Six months postoperative: (A) external finding and (B) intra-oral finding.

suffice for small-sized defects, while larger defects may require a local, regional, or free flap[19,20]. Medium-sized defects, typically 2–5 cm, can be particularly challenging[19–21]. Various local flaps have been suggested as options for reconstruction, such as the buccal fat pad flap, facial artery musculomucosal flap (FAMM), buccinator musculomucosal flap, masseter muscle flap, tongue flap, and palatal flap[19,21–23]. Each technique offered distinctive benefits and a high success rate[19,21–23]. According to a treatment algorithm presented in 2018[19], the buccal fat pad was considered the most appropriate option for buccal mucosa reconstruction. Nevertheless, epithelialization of the buccal fat pad following surgery typically requires approximately 3 weeks to 2 months for complete healing[19,21,22]. There have been reports of minor complications such as partial flap necrosis, wound dehiscence, local infection, and limited mouth opening[19,21,22]. In addition, a potential solution for a medium-sized defect in the buccal mucosa was using a mucosal flap from the floor of the mouth[20]. However, this option was only suitable if the defect is situated below the occlusion line, and extraction of relevant teeth may be necessary[20].

The reconstruction ideal would be to replace the damaged tissue with a similar one, as it offers original properties[4], and has the same pliability and bulkiness. As a result, a healthy buccal mucosa flap was deemed suitable for this situation for defects up to 4 cm in size. This option was also less time-consuming, with a quick wound-healing process and no restricted mouth opening, making it an appropriate choice for reconstructive procedures. The flap with anterior-based random vascular supply can be restored to both buccal mucosa and lip defects simultaneously. Although a minor complication was observed in small wound dehiscence, no evidence of wound infection, flap necrosis, bleeding, or hematoma was reported. Furthermore, a thorough follow-up of the primary site and neck can be achieved, and both functional and facial esthetic outcomes were accomplished.

The practical consideration observed in the case study was the importance of selecting a biopsy location that avoids areas of inflammation and necrosis to ensure accurate diagnosis. This issue can be prevented by improving communication between pathologists and physicians to ensure precise diagnosis and treatment decisions. Furthermore, the buccal advancement flap was a suitable option for medium-sized buccal defects and could be performed by a general otolaryngologist without the need for special instruments, which were available at secondary care hospitals.

The limitation was the initial diagnosis was unclear, requiring further investigation, including immunohistochemistry staining for better accuracy. Insufficient intraoperative pathological settings may have caused the removal of healthy tissue beyond the necessary margins.

Conclusion

In medium-sized buccal mucosa defects, the buccal advancement flap is a highly effective treatment option that produces favorable functional and facial esthetic outcomes. This technique is straightforward, involves only one stage, and results in minimal complications and good healing. Additionally, it is possible to use this method for reconstructing buccal defects up to 4 cm in size, making it a valuable option.

Ethical approval

This issue has been approved by the ethics committee of the author's institute, with the study code 66-02-20, COA NO 199.

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

C.S. and K.Y.: conception of the work, drafting and revising the work, final approval of the version to be published, and agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Conflicts of interest disclosure

There are no conflicts of interest.

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Data availability statement

The research data used during the current study are available from the corresponding author upon reasonable request.

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