Case Reports

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Monomorphic Adenoma of Minor Salivary Gland: An Unusual Diagnostic Challenge

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Abstract

Objective: To describe a rare case of monomorphic adenoma of the minor salivary gland.

Clinical Presentation and Intervention: A 50-year-old patient presented with a painless swelling of 2-3 years duration in the chin region, appearing large permeative destruction lesion involving symphysis-menti and adjacent bodies of mandible with soft tissue component infiltrating into right sublingual and sub-mental space in MRI imaging.

Histological findings were suggestive of a monomorphic adenoma.

Treatment: A complete surgical enucleation of the lobular tumor with an additional zone of adjacent normal tissue was performed. No recurrence was observed after a 6-month follow-up.

Conclusion: This case showed the possibility of minor salivary gland tumors and highlighted the need to consider them when making a differential diagnosis of swellings located in the sub-mental region.

Keywords: Monomorphic adenoma, Salivary gland tumor, Surgical enucleation.

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INTRODUCTION

A salivary gland neoplasm constitutes less than 1% of all tumors and 3-5% of head and neck tumors. Minor salivary gland tumors are rare, comprising 10-15% of all salivary gland neoplasms, primarily situated in various locations: Palate (50%), lips (15%), cheek mucosa (12%), tongue (5%), and floor of the mouth (5%).[1,2] Monomorphic adenoma is an uncommon benign epithelial tumor of the salivary gland, representing 1-2% of all salivary gland tumors.[3] Classification of salivary gland monomorphic adenomas involves various principles, such as cell type, cellular arrangement, and embryologic development, leading to a diverse group of tumors. Histologically, four characteristic patterns are described: solid, trabecular, tubular, and membranous.[4] Monomorphic adenomas are typically slow-growing, encapsulated tumors, firm in consistency, mobile, and painless, often appearing superficially within the glandular body with a brownish hue and usually not exceeding 3 cm in diameter.[5,6] The clinical presentation of monomorphic adenoma may resemble that of a mucocele of the oral mucosa, with the former more commonly appearing in the upper lip of older individuals and the latter in the lower lip of younger individuals.[7]

CASE REPORT

A 50-year-old patient presented to the department with a complaint of a large swelling over the chin region persisting for 2 to 3 years. The patient described the swelling as non-tender, gradually increasing in size over time, leading to facial asymmetry. Additionally, there was a history of numbness or paresthesia of the lower lip. Apart from these symptoms, the patient was in good health.

Upon examination, an oval-shaped swelling measuring 5x6 cm was observed extra-orally in the sub-mental region, extending medio-laterally from one commissure of the lip to the other. Superiorly, it extended 1.5 cm below the lower border of the lower lip, and inferiorly it reached 2 cm beyond the sub-mental region. (*Fig. 1*) The swelling was mobile, firm in consistency, and not fixed to the underlying structures. There was no tenderness, fluctuation, or localized increase in temperature. Intra-orally, a well-defined solitary swelling measuring up to 4x2 cm was observed in the labial vestibule, specifically in the region of teeth 31, 32, 41, and 42, leading to the obliteration of the labial vestibule. The overlying mucosa appeared intact without any color differentiation from the surrounding mucosa. (*Fig. 2*)

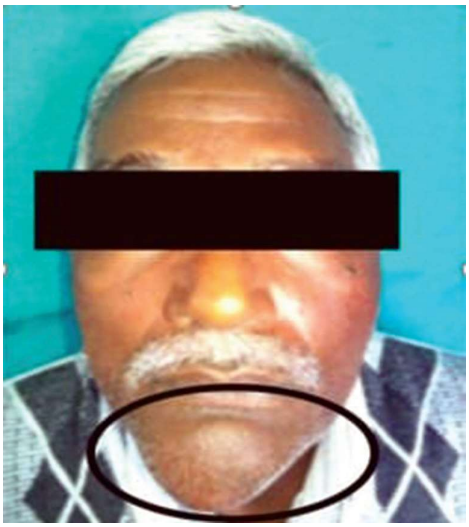

Fig. 1: Extra oral picture showing sub-mental swelling

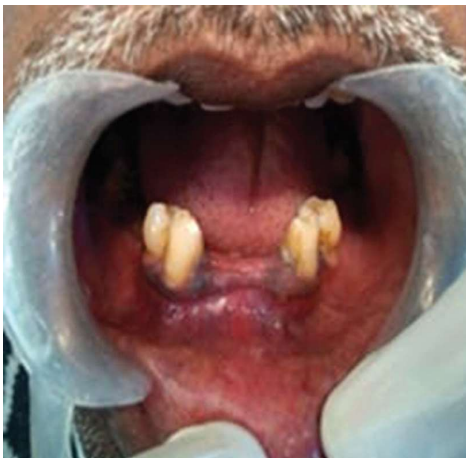

Fig. 2: Intraoral picture showing diffuse swelling in labial vestibule

Based on the patient's history and clinical examination, a provisional diagnosis of a benign neoplasm involving the labial vestibule region was made, with a differential diagnosis including odontogenic myxoma, central giant cell granuloma, monomorphic adenoma, pleomorphic adenoma, and adenoid cystic carcinoma. Radiographic examinations including OPG and lateral oblique view of the mandible revealed heterogenous areas with irregular margins and erosion of the inferior margin of the mandible in the symphysis-menti

area. (*Fig. 3, 7.4*)

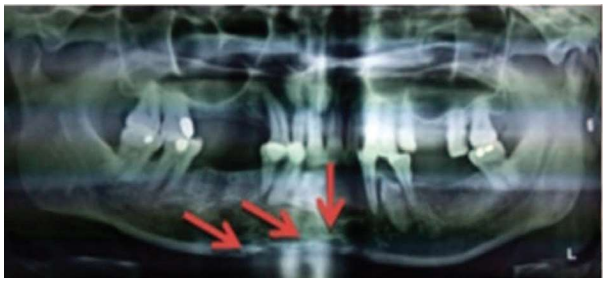

Fig. 3: OPG showing heterogeneous lytic areas and erosion of inferior margin of the mandible

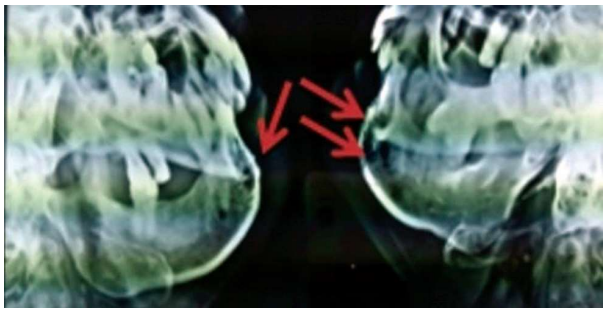

Fig. 4: Mandibular lateral oblique view showing heterogenous lytic areas and erosion of inferior margin of the mandible

The MRI report indicated a large permeative destruction lesion involving the symphysis-menti and adjacent bodies of the mandible on both sides, with cortical breakdown and infiltration into the right sublingual and sub-mental space muscles and soft tissue. The FNAC report of the submental swelling showed clusters of small monomorphic cells. Subsequently, a biopsy of the sub-mental swelling was performed under local anesthesia, and the histological features were suggestive of monomorphic salivary adenoma.

A complete surgical enucleation of the lobular tumor, along with an additional zone of adjacent normal tissue, was performed under local anesthesia. After a 6-month follow-up, no recurrence was observed; however, the patient was lost to further follow-up.

DISCUSSION

The minor salivary glands are comprised of 600-1000 small independent glands distributed throughout the oral cavity, palatine tonsils, pharynx, and larynx.[8] Bertrandi was credited with the concept of salivary gland tumor exegesis in 1802.[9] These glands are the second most common site of involvement, accounting for 28.5% of all tumors.[10]. Monomorphic adenoma is an uncommon benign epithelial tumor of the salivary gland, representing 1-2% of all salivary gland tumors, rendering cases like ours rare. Typically, it afflicts individuals between their fifth and seventh decades, which aligns with our findings in this 50-year-old male patient.

The etiological factors contributing to general salivary gland neoplasms are not precisely defined. However, some research suggests associations with factors such as radiation therapy in low doses, smoking, Epstein-Bar virus infection, genetic predisposition, sunlight exposure, vitamin A deficiency, and chemotherapy. These tumors usually manifest as slow-growing asymptomatic masses, posing challenges in diagnosis, management, and predicting the clinical course of the disease.

To achieve a definitive diagnosis and differentiate this neoplasm from others in the differential diagnosis, fine-needle aspiration cytology (FNAC) and histopathological analysis played pivotal roles, as was the case in our patient.

The optimal management approach for monomorphic adenomas of the minor salivary glands involves wide surgical excision, including a margin of normal tissue. The extent of excision varies depending on the tumor's location, size, and histology. Favorable prognosis is generally achieved after wide excision. However, regular follow-up is imperative due to the documented risk of malignant transformation and recurrence noted in the literature.

In our case report, wide excision was performed, followed by a 6-month postoperative follow-up period during which no recurrence was observed. Unfortunately, the patient was lost to further follow-up.

CONCLUSION

Wide surgical excision remains the standard treatment modality for the management of minor salivary gland tumors. This case underscores the importance of considering minor salivary gland tumors in the differential diagnosis of submental swellings and highlights the efficacy of surgical management in achieving favorable outcomes.

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