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Pleomorphic Adenoma Arising from Accessory Parotid Gland - A Diagnostic Dilemma

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INTRODUCTION

Less than 3% of all head and neck tumours have salivary gland aetiology. They are most commonly associated with parotid gland. Submandibular salivary gland and minor salivary gland have less frequent association. Molar glands are a small group of secretory glands situated between the masseter and buccinator muscles and beneath the mucosa around the distal extremity of the duct of the parotid gland. Their ducts open into the mouth opposite the last molar tooth. Tumours arising from these minor glands often presents as mid cheek swelling. The differential diagnoses for mid cheek swelling are many, with salivary gland tumours being most common. Malignant salivary gland tumours are more commonly seen to be associated with minor salivary glands. Benign minor salivary gland generally presents as a symptom-free mass and often has the potential to attain huge size. Surgical excision is the standard for treatment. Tumour recurrence is often seen due to incomplete removal. Malignant transformation has also been reported.

Pleomorphic adenomas are most commonly found in major salivary glands wherein they are seen arising from myoepithelial and intercalated duct cells. However, tumours arising from minor salivary gland are rarely seen. 'Molar glands' are a small group of secretory glands situated between the masseter and buccinator muscles and beneath the mucosa around the distal extremity of the duct of the parotid gland. Their ducts open into the mouth opposite the last molar tooth. This case report elaborates a rare lesion in a 38-year-old female, which was diagnosed as pleomorphic adenoma arising from molar gland. A 1.6 cm tumour which was well defined and circumscribed, presenting as submucosal nodule characterized by slow indolent growth and firm on palpation was seen in the mid cheek region. Following complete excision, the histopathological evaluation suggested pleomorphic adenoma.

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PRESENTATION OF CASE

A 38-year-old female presented to the outpatient clinic of our center with a painless mass in the left mid-cheek region. The patient reported that the mass appeared four years earlier with gradually increase in size. Past medical history was unremarkable. The mass was lying submucosally in the left cheek, (figure 1) it was hard on palpation and non-adherent to the surrounding tissues. Ipsilateral facial nerve function was normal. Magnetic resonance imaging of the facial skull with gadolinium contrast revealed a well-demarcated, oval-shaped tumour in the anterior border of the left masseter muscle with high intensity signal in T2-weighted images which was markedly enhanced by contrast. Maximum tumour diameter of the neoplasm was 1.6 cm. (figure 2)

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Surgical removal of the tumour was planned. A linear incision was made over the left buccal mucosa avoiding the left t parotid duct opening. Intraoperatively, the tumour was dissected free of the surrounding tissues and removed in total without any residual morbidity. Histopathological examination of the tumour reported as pleomorphic adenoma of the minor salivary gland. Following the surgery, the facial nerve function was normal postoperatively and the patient was discharged two days after surgery. Patient recovery period was uneventful.



Figure 1.
Submucosal Growth in the
Left Cheek Region Abutting
the Parotid Ductal Opening

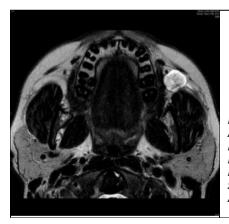


Figure 2.
Axial T2- Weighted
MRI Image, Showing
Hyperintense Well
Demarcated Oval
Shaped Tumour
Anterior to Masseter

DISCUSSION

Tumours of mid-anterior cheek presents as various diagnostic possibilities. Most common these tumours relate as lymphadenopathy, other reported conditions include hemangioma, fibroma, lipoma, schwannoma or neurofibroma. Primary parotid duct tumours, and accessory parotid gland lesions holds an important differential for mid cheek swelling due to its anatomical association with the area. 22% of all salivary gland neoplasms have minor salivary gland aetiology.¹ Majority of these tumours are malignant with benign tumours accounting for only 18%. Of all benign tumours originating for minor salivary gland pleomorphic adenoma is the

commonest.¹ Most commonly pleomorphic adenoma of the minor salivary gland is seen arising from palate followed by lip, buccal mucosa and floor of mouth less frequently in tongue, tonsil and retromolar region.¹-³ Submucosal buccal minor salivary glands are most commonly found to be the etiology for mid cheek buccal Pleomorphic adenomas.⁴ Minor salivary glands in the buccal space, present deep to the buccinator has rarely been reported in the literature. These glands are referred to as the buccal or molar glands and these glands usually drain opposite the last maxillary molar teeth with their ducts traversing through the buccinator.⁵

Most of the minor salivary gland tumours present as submucosal mass or nodule. The rate of growth of the neoplasm is usually slow and the tumour is often insidious in onset. 50 patients with minor salivary gland tumours of the lip and buccal mucosa were studied and the median duration for the symptoms to appear prior to diagnosis was found to be five months for buccal tumours.6 Fine-needle aspirate cytology (FNAC) is an essential tool for evaluation of a mid-cheek mass were a tissue biopsy can sometimes be challenging due to the anatomical orientation of the tumour. CT and MRI scans are often helpful in providing information regarding anatomic relationships and also in planning the FNAC and future surgery.^{7,8} Smith et al⁷ emphasized on sialography in addition to CT for differentiating intrinsic parotid gland tumour from extrinsic tumours. MRI scan holds importance in diagnosing mid cheek tumours. It helps in providing information regarding the extent of local spread as well as sometimes gives critical information on the type of tumour. Intact fat plane in a MRI is an important distinguishing feature between malignant and benign tumours. It also helps in delineating the extent of tumour external to buccinator and association with parotid and Stenson's duct. MRI shows characteristic hypointensity on T1 weighted image and is heterogeneously hyperintense on T2 weighted image with multiple well-defined areas of low cellularity within without any invasion to adjacent tissues Histopathologically, pleomorphic adenoma has a complex morphological arrangement of, epithelial and myoepithelial cells presenting varied pattern within a mucopolysaccharide stroma. Capsule of the tumour is essentially formed as a result surrounding connective tissue fibrosis therefore sometimes referred to as "false" capsule.9

The treatment of pleomorphic adenoma is surgical excision. Spiro reported a series of 1342 patients with benign parotid neoplasms in which the recurrence rate was found to be 7%. They had further reported 6% recurrence rate in patients with other benign minor salivary gland tumours.¹ Resection of the tumour with emphasis on clear margin is paramount for preventing any local recurrence.¹¹ Due to the presence of capsule the recurrence rates are less. "Dehiscence" with presence of false capsule can sometimes compromise the margin of resection leading to recurrence, henceforth adequate margin of gross normal surrounding tissue is necessary for adequate removal of the tumour. Tumour spillage due to rupture of the capsule is another reason for increased risk of recurrence, so careful dissection is advised.

CONCLUSIONS

Mid-cheek lesion proposes a high chance for being a minor salivary gland tumour. Thorough understanding of the

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anatomical location of this lesion and assimilating the clinical diagnosis with radiologic investigation in the form of an MRI helps in reaching the diagnosis and deciding on surgical plan with least morbidity.

REFERENCES

- [1] Spiro RH. Salivary neoplasms: overview of a 35-year experience with 2,807 patients. Head Neck Surgery 1986;8(3):177-84.
- [2] Wang D, Li Y, He H, et al. Intraoral minor salivary gland tumours in a Chinese population: a retrospective study on 737 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104(1):94-100.
- [3] Cohen MA. Pleomorphic adenoma of the cheek. Int J Oral Maxillofac Surg 1986;15(6):777-9.
- [4] Torabinia N, Khalesi S. Clinicopathological study of 229 cases of salivary gland tumours in Isfahan population. Dent Res J (Isfahan) 2014;11(5):559-63.

[5] Pick TP, Howden R. Gray's Anatomy. 15th edn. London: Chancellor Press 1985: p. 853.

- [6] Weber RS, Palmer JM, El-Naggar A, et al. Byers Minor salivary gland tumours of the lip and buccal mucosa. Laryngoscope 1989;99(1):6-9.
- [7] Smith JR, King WW, Tang WY, et al. Differentiating tumours of the deep and superficial lobed of the parotid gland by computed tomographic sialography. Clin Radiol 1987;38(4):345-9.
- [8] Toida M, Shimokawa K, Makita H, et al. Intraoral minor salivary gland tumours: a clinicopathological study of 82 cases. Int J Oral Maxillofac Surg 2005;34(5):528-32.
- [9] De Las Casas LE, Hoerl HD, Oberley TD, et al. Myoepithelioma presenting as a midline cystic tongue lesion: cytology, histology, ancillary studies and differential diagnosis. Diagn Cytopathol 2001;24(6):403-7.
- [10] Mendenhall WM, Mendenhall CM, Werning JW, et al. Salivary gland pleomorphic adenoma. Am J Clin Oncol 2008;31(1):95-9.