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UNCOMMON SALIVARY GLAND TUMORS: A CASE SERIES

General Surgery

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ABSTRACT

Salivary gland tumors are one of the rare entities having the annual incidence rate of 0.5 to 2 per 100,000 persons[7]. Majority of them are benign, only 20% cases are malignant. They usually present as painless mass in the oral cavity or close to their anatomical location. The main challenge is to differentiate between the benign and the malignant ones. Thus, various imaging modalities are used. Investigations such as fine needle aspiration cytology and MRI scans provide some useful information, but most cases will require tissue diagnosis to come to a definitive conclusion. Hence, biopsy or histopathological study of the surgically excised tissue is the gold standard for diagnosis. Benign tumour and early low-grade malignancies can be adequately treated with surgery alone, while more advanced and high-grade tumour with regional lymph node metastasis will require postoperative radiotherapy[11]. The role of chemotherapy remains largely palliative. In this article, we have presented a series of cases with uncommon tumors involving the various salivary glands.

KEYWORDS

Myoepithelioma, Mucoepidermoid Carcinoma, Pleomorphic Adenoma, Salivary Gland Tumors.

INTRODUCTION:

Neoplasms that arise in the salivary glands, namely the Parotid gland, the Submandibular gland, the Sublingual gland and the minor salivary glands, are relatively rare. In spite of that, they represent a wide variety of both benign and malignant histologic subtypes. Approximately 70% of the salivary gland tumors affect parotid gland with the submandibular gland being affected in 5-10% of the cases, sublingual gland in 1% and minor glands in 5-15% of the cases[1,2]. Some 80 – 90% of tumors of the Parotid are benign, and the most common being Pleomorphic adenoma[4,5]. For Submandibular gland most common tumor is Adenocystic carcinoma (40%)[3,5].Tumors involving Sublingual and the minor salivary glands are extremely rare and are almost always (90%) malignant[5]. The World Health Organization (WHO) in 2005 recognized 24 different malignant salivary gland cancers. The most common histologies include mucoepidermoid carcinoma (MEC), acinic cell carcinoma (ACC), adenoid cystic carcinoma (AdCC), carcinoma ex-pleomorphic adenoma (CExPA), and adenocarcinoma[3]. Here we present a series of uncommon salivary gland tumors involving the Parotid, the Submandibular and the Minor salivary glands.

CASE PRESENTATION:

Our first patient was a 27-year-old female who presented to the General Surgery OPD, with the chief complaint of a painless swelling in the right side of cheek for past 2 years. It was a pea-shaped, firm and mobile swelling to start with, which increased in size over the past 6 months. There was no history of pain associated with salivation or intake of food or dryness of mouth. Neither there was any allergic history to any food or medication, nor there was any history of addiction to tobacco or other substances in any form. On examination [Fig. 1], a single, globular mass, approximately of 3 x 3 cm, was present in the right side of cheek, just lateral to the right nasolabial fold. The swelling had a smooth surface, regular margin, well defined border, and it was freely mobile in the subcutaneous plain. Overlying skin was normal, no visible pulsation or deviation of angle of mouth or dribbling of saliva, or loss of any facial expression.

On Ultrasonography, a well-defined homogenous hypoechoic solid mass, measuring approximately 24 x 17 mm, was noted in the submucosal level deep to the buccinator muscle. The mass had a significant internal vascularity, possibly represented a minor salivary gland neoplasm. The parotid gland and duct were normal. There was no evidence of parotid duct dilatation. Fine needle aspiration cytology (FNAC) of the swelling revealed Monolayer sheets of myoepithelial cells, few clusters of duct epithelial cells with extracellular chondromyxoid matrix in a haemorrhagic background. Overall features were suggestive of Pleomorphic Salivary Adenoma CAT II in the Milan system of reporting salivary gland cytology. The patient underwent a wide local excision of the swelling via transoral approach under general anesthesia. The procedure was uneventful. A well circumscribed firm encapsulated mass measuring 3.5 x 2.5 x 2 cm was excised from the oral cavity of the right side. The cut surface appeared to be glistening white in colour with occasional bosselations. The specimen was sent for histopathological examination, which revealed **Myoepithelioma**, arising from one of the Minor Salivary Glands.

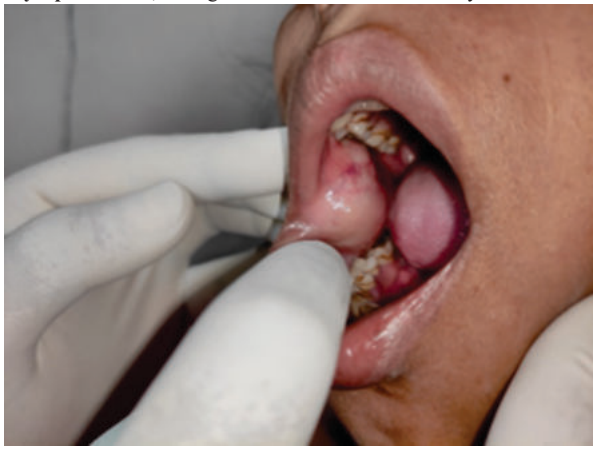

Fig. 1: Right sided intra-oral swelling

The second case was of a 25-year-old female patient presenting to the OPD with swelling behind right ear below mastoid for 5 years [Fig. 2]. It was a painless swelling, which gradually increased in size over the past few years. There was no history of pain associated with salivation or intake of food or dryness of mouth. There was no history of deviation of angle of mouth or any facial abnormality. The patient had no history of addiction in any form. On examination, it was a soft to firm swelling, measuring approximately 4 x 2 cm, on the right side of the cheek, just below the ear lobule. The swelling had a smooth margin, well defined borders, and it had restricted mobility. The overlying skin was normal. There was no deviation of angle of mouth or dribbling of saliva, or loss of any facial expression. Ultrasound of the mass revealed, one complex cystic SOL measuring 3.8 x 1.8 cm, in close relation to the right parotid gland, with internal solid component. Fine needle aspiration cytology (FNAC) of the swelling revealed the possibility of Pleomorphic salivary adenoma of the Parotid gland. The patient was planned for superficial parotidectomy under general anesthesia. After dissecting the tissue, the tumour was found to be arising from the posterior margin of the right parotid gland. Only the

mass was excised as the gland was found to be normal. Post-operative histopathology of the specimen revealed **Low-grade Mucoepidermoid carcinoma** of the Right Parotid Gland.

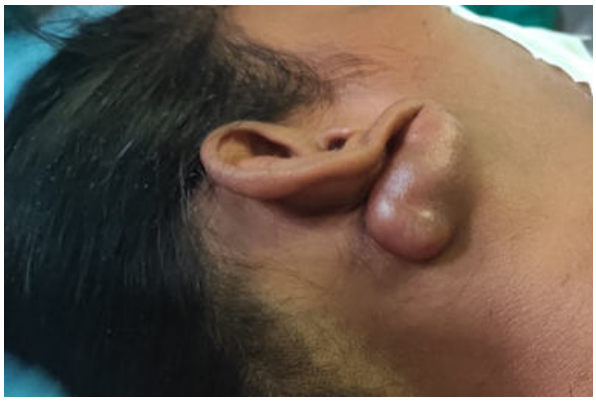

Fig. 2: Right postauricular swelling

On the third occasion, we had this 52-year-old female patient attending our OPD with the complaint of a swelling in the right side of the neck, below the lower jaw, for last 6 months [Fig. 3]. It was a painless, non-mobile mass which was insidious in origin with gradually progressing in nature. There was no history of pain associated with salivation or intake of food or dryness of mouth. The patient had no history of addiction in any form. On examination, it was a firm bosselated mass in the right submandibular region, measuring approximately 4 x 3 cm, having a regular margin and a well-defined border. The mass had restricted mobility. Intraoral examination revealed no reduced salivation from right Wharton's duct. Ultrasonography of the mass described the lesion as a hypoechoic SOL, measuring 3.5 x 2.3 cm, with internal vascularities, arising from the right Submandibular gland. Fine needle aspiration cytology (FNAC) of the swelling addressed it as a possibility of Pleomorphic adenoma of the salivary gland. The patient was taken up for excision of the mass under general anesthesia. A greyish white tissue was excised and sent for histopathological examination. The histopathology suggested it to be **Pleomorphic Adenoma** of the Right Submandibular Gland.

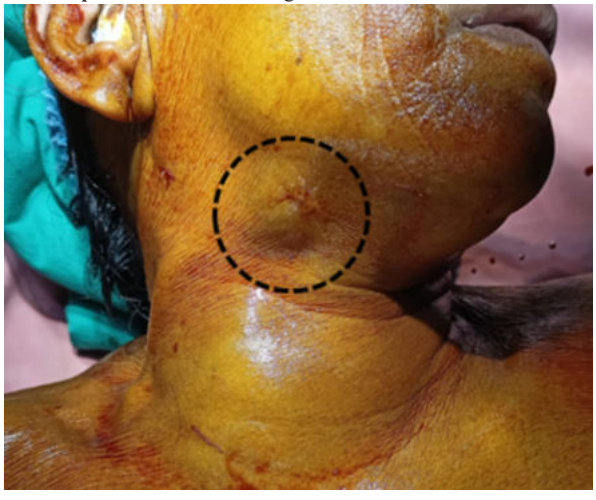

Fig.3: Right submandibular gland swelling

DISCUSSION:

The majority of salivary gland neoplasms are benign and only 20% are malignant[6]. The annual incidence of salivary gland cancers ranges from 0.5 to 2 per 100,000 in different parts of the world[7]. The sex distribution for salivary gland cancers is almost equal, and the majority of the cases arise in the sixth decade[8]. Tumors can occur in both the major and minor salivary glands. 80% of major salivary gland tumour occur in the parotid glands, while most minor salivary tumour are located in the palate[9]. Whilst most other head and neck cancers are strongly related to smoking and drinking, these do not play a role in the salivary glands. Some studies have found that a diet rich in vitamin C and low in cholesterol may be effective in preventing salivary gland cancer[10]. Parotid or Submandibular gland tumors often present as an enlarging mass. They occasionally can cause neurological symptoms such as facial nerve paralysis or pain, if they are malignant[2]. Minor salivary gland tumour presents as a submucosal intraoral mass[11]. Ultrasound is the first non-invasive option for evaluating major salivary gland tumors. It can help to localize tumors; distinguish solid masses from cystic collections, and help guide fine-needle aspiration biopsy. Heterogenous echogenicity, local invasion, poorly-defined margins, and lymphadenopathy are sonographic signs of malignancy[12,13]. Conventional CT can evaluate tumor extent, bony infiltration, and lymphadenopathy. However, it is limited by the dental artifact and has a poor soft-tissue resolution[6]. For lesions in the deep parotid lobe, sublingual glands, and minor salivary glands, MRI is recommended to assess tumor extent, soft tissue invasion, and nerve involvement. MRI can be performed to detect the facial nerve branches and their interface with surrounding soft tissue for surgical planning. MRI has a higher sensitivity and specificity than CT in detecting perineural spread[14]. The role of PET is to detect locoregional and distant metastasis. In demonstrating tumor extension, nodal involvement, local recurrence, and distant metastasis, PET scan is more accurate than conventional CT scan[15,16]. However accurate the imaging techniques may be, they cannot distinguish between benign and malignant lesions completely. Hence the tissue sampling is always gold standard for confirmatory diagnosis. An incisional biopsy is not recommended for parotid lesions due to the risk of damage to the facial nerve and the possibility of tumor-seeding. Hence, ultrasound-guided fine-needle aspiration (FNA) is preferred[17]. However, FNA may falter in its ability to determine the specific malignant subtype and tumor grade[18]. After a definitive diagnosis is made, surgical excision with negative margins is the mainstay of treatment for all salivary gland malignancies. The extent of surgery and the need for neck dissection or adjuvant radiotherapy will be dependent on the subtype, grade, and stage of the malignancy[6].

CONCLUSION:

Salivary neoplasms present with varying histologies and severities, often requiring multimodal therapy involving surgery, radiation, and chemotherapy. In the cases we have described above, we had encountered uncommon types of salivary gland tumors. While Pleomorphic adenoma is common in Parotid gland, we encountered Mucoepidermoid carcinoma of the parotid. This has an incidence rate of 2.3 per 1,000,000[19,20]. For Submandibular gland, Adenocystic carcinoma is the most common variety, but our patient had Pleomorphic Adenoma of the gland. On the other hand, Myoepithelioma is one of the rare salivary gland tumors, comprising of only 1 – 1.5% of the cases[21]. The Parotid is the most commonly affected gland, (~ 40%), whereas, the minor salivary glands are involved less frequently (~ 21%)[22,23]. Thus, our patient had one of the rare tumors of the minor salivary gland. In all of the three cases, the subjects were followed-up for one year, in which none of them had any signs of any recurrence or any post-operative complications.

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