PLEPMORPHIC ADENOMA: A RARE SIGHTING OF A COMMON TUMOUR

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Abstract

Background: Salivary gland tumours comprise of 2-3% of head and neck tumours. Pleomorphic adenoma is the most common among all the major and minor salivary gland tumours.

Case Report and Follow up: We present a case of pleomorphic adenoma in the tongue, which was clinically diagnosed as Irritational fibroma due to its site of presentation and histologically was diagnosed as pleomorphic adenoma. The case has been followed up for postoperatively for recurrence.

Conclusion: Pleomorphic adenoma of minor salivary gland is a tumour of rare occurrence and a diagnosis should be made carefully lest a major salivary gland be resected. High index of suspicion and an adequate clearance of the tumour with a cuff of surrounding dispensable normal tissues is the key to successful treatment of such tumours.

Kerwords: Irritational Fibroma, Pleomorphic Adenoma, Tongue.

Introduction

Pleomorphic adenoma (PA) accounts for a total of 40% to 70% of all major and minor salivary gland tumours making it the most common.[1,2] Generally considered to be benign, they present with several histological features. The high recurrence rate and frequent malignant conversion form its main characteristics. For most cases, the only clinical sign is an asymptomatic painless swelling with a slow rate of growth. Microscopically, PA is characterised by variable, diverse structuring histological patterns consisting of fibrous, hyaline, myxoid, cartilaginous and osseous areas, which are differentiated by myoepithelial cells. PA's prime site of involvement is the parotid gland, and the minor salivary glands are the least involved. Among the minor salivary glands, PA occurrence for tongue is reported to be very rare. Certain studies have shown that PA in the minor salivary gland mostly occurs during the second decade of life, only 5 to 10% affect patients aged 20 years and under,[2-4] while others studies show that the mean age for males was in the 3rd – 4th decade of life.[5]

Hence, the purpose of this article is to report a case of pleomorphic adenoma with the characteristic histologic features in an unusual location.

Case report

A 55-year-old female patient reported to the Department of Oral and Maxillofacial Surgery, with a complaint of a growth on the right side of the tongue that she noticed for 15 days. There was a sudden onset with no increase in size since the time noted and she did not experience difficulty in any function. No history of bleeding or any discharge from the growth or pain and sensory changes associated.

Physical examination intra orally, revealed a solitary nodular growth measuring about 1.5cm x 1cm in dimensions in the right lateral border of the tongue adjacent to 46, 47 where the tongue rested. [Figure 1] Surface of the growth appeared lobulated and erythematous with surrounding mucosa appearing normal. [Figure 2]

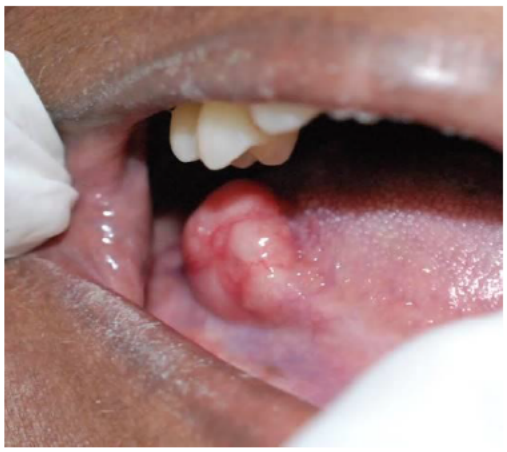

Figure 1: Intraoral photograph of the swelling on the right lateral border of tongue.

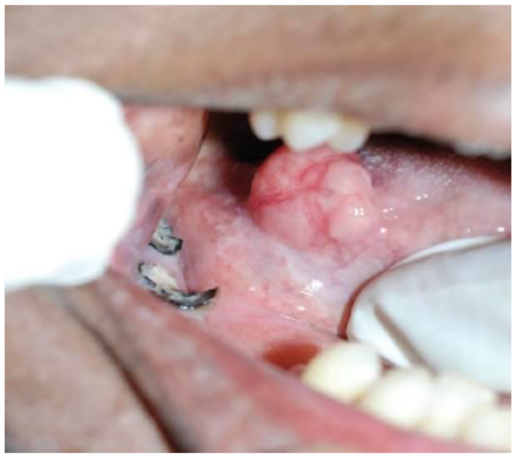

Figure 2: Intraoral photograph of the swelling adjacent to grossly decayed 46, 47.

On palpation, size and extent were confirmed. Growth was non-tender, firm in consistency, sessile, non-fluctuant, non-reducible, compressible and non-pulsatile. No evidence of blanching was observed on digital pressure. There was no history of bleeding on palpation. Hard tissue examination revealed root stumps in relation to 17 & 45 (tooth number). Grossly decayed teeth with sharp margins in relation to 47

and 48 were noticed. Extra orally, no gross abnormality was detected. Investigations we ordered included Orthopantomogram, Fine needle aspiration cytology (FNAC) and an excisional biopsy. Routine hematologic investigations and urine analyses were normal. Panoramic radiography revealed no abnormality. Based on clinical findings and examination a provisional diagnosis of Irritational Fibroma on the right lateral border of tongue was made. Subsequently, an excisional biopsy of the mass was performed under local anaesthesia (2% lignocaine with 1:80000 adrenaline). A 5mm margin of clinically healthy tissue was included and the depth of the excision was up to the muscle layer of the tongue. The lesion was non-adherent to the surrounding mucosa. The tissue was firm of palpation. The specimen was marked and sent for routine histopathological examination. The histologic features of the mass were consistent with the cellular variant of PA. [Figure 3]

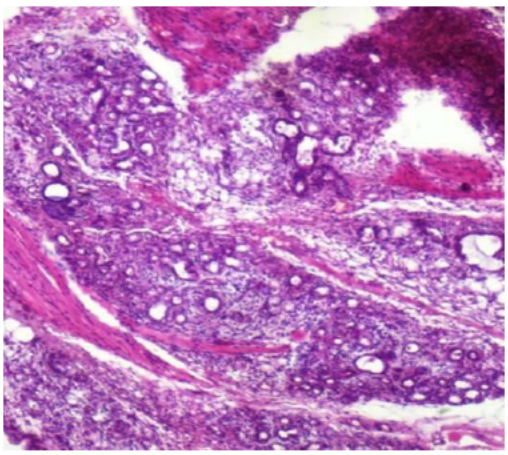

Figure 3: Photo micrograph (40x) showing fibrous, hyaline and myxoid components which are differentiated by myoepithelial cells.

As the histopathology report differed from our provisional diagnosis of Irritational Fibroma, a secondary surgical procedure of wider excision was done using the carbon dioxide laser. This was done to ensure all margins are free of tumour and the specimen was marked and sent for routine histopathological examination. The secondary excisional specimen had no evidence of lesional tissue in this section. The report revealed no evidence of pathology in the given sections and that all the margins were the free of tumour. The postoperative period was uneventful and the patient has been tumour-free till date.

Discussion

Tumours of the salivary glands are of intricate origin consisting of epithelial and connective tissues. The majority of salivary gland neoplasms are benign with pleomorphic adenomas being the most common. Pleomorphic adenoma is more prevalent in the 4th - 6th decade of life and is more common in females than males. Pleomorphic adenomas are the most common types of salivary tumours (40-60%) and mostly occur in the parotid gland, which accounts for 60-80% of Pleomorphic adenomas. Pleomorphic adenoma is the most common neoplasm of the minor salivary glands (39% of cases) but compared with tumours of the major salivary glands, a greater proportion of minor salivary gland tumours are malignant.[6]

Pleomorphic adenomas arising from oral minor salivary glands account for approximately 10% of pleomorphic adenomas with the posterior hard palate and anterior soft palate being the most common sites.[7] "Yoshihara and Suzuki found that the majority of pleomorphic adenomas involved the palate, followed by the lips."[8] Involvement of the tongue was extremely rare. When they occur on the tongue, pleomorphic adenomas are most commonly seen in the posterior, followed by anterior, and rarely in the lateral-lingual gland of the tongue (Ebner's gland).[9] Malignant tumours involve the tongue more frequently than their benign counterparts. Malignant tumours such as adenocarcinoma, adenoid cystic carcinoma, and mucoepidermoid carcinoma involve the tongue more frequently.[8,10,11]

Of 1320 tumours reviewed by Chaudhary *et al*., the base of the tongue was the site of origin in only 1% of all intraoral minor salivary gland tumours.[12]

Pleomorphic adenoma is a benign tumour with slow growth; it seems not to change for many years and expresses itself simply as a non-painful mass.[13] They are usually well demarcated or encapsulated but extension of tumour into the capsule is a common feature and sometimes lobules of tumour may appear to be completely separated from the main tumour mass. PA of intraoral accessory glands seldom is allowed to attain a size greater than 1-2 cm in diameter. Because this tumour causes the patient difficulties in mastication, talking and breathing, it is detected and treated earlier than other tumours of major glands.

Differential diagnosis we considered due to the clinical presentation and localization of the lesion, included mucocele, reactive lesions such as giant cell fibroma or focal fibrous hyperplasia, lipoma, granular cell myoblastoma, neurofibroma, neurilemmoma, vascular leiomyoma, and benign salivary gland neoplasm in the differential diagnosis. The diagnosis of Irritational fibroma was obtained since the lesion rested adjacent to grossly decayed teeth with sharp margins.

Treatment for pleomorphic adenoma is primarily surgical. Although these tumours are well encapsulated, resection of the tumour with an adequate margin is essential to avoid recurrence. Enucleation as a primary modality increases the chances of recurrence, thus excision is always done with safe margin around.

Approximately 20-45% of pleomorphic adenomas recur despite surgical excision and 2-9% will degenerate to malignant tumours. The risk of recurrence is increased if the capsule is torn. In cases of the aged, malignant change in benign tumours of the minor salivary gland, that is carcinoma-ex pleomorphic adenoma must be taken into consideration. The follow-up of patients with salivary gland

tumours should be long due to the possibility of late recurrences.[3,14]

Follow Up

The patient was reviewed post-incisional biopsy for healing of the wounds edema, pain and bleeding. Following the histopathological diagnosis, excicion of the lesion was done using Carbon Dioxide laser. In the post operative period we observed better healing, edema and pain subsided in 2 weeks. Patient has been followed up for every week in the first month and every three months for the past two years. There is no evidence of recurrence.

Conclusion

The salivary glands may show a diverse range of lesions presenting a challenge to even the most experienced clinician and pathologist. Pleomorphic adenoma of minor salivary gland is a tumour of rare occurrence and a diagnosis should be made carefully lest a major salivary gland be resected. High index of suspicion and an adequate clearance of the tumour with a cuff of surrounding dispensable normal tissues is the key to successful treatment of such tumours.

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