CASE REPORT

Pleomorpic Adenoma of Infratemporal Fossa Co-Existing with Actinomycosis: A Rare Case Report with Review of Literature.

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Abstract

Introduction: Salivary gland tumors account for about 3% of head and neck tumors and the majority are benign in nature. Among these, the most common pathological type is pleomorphic adenoma also called the mixed tumor. It is a kind of tumor containing glandular tissue, myxoid and cartilage-like tissue. As the structure of the tissue is diverse, it is called a "mixed tumor." Actinomycosis is a chronic suppurative bacterial infection caused by *Actinomyces israelii*. In this case report, we are presenting a rare co-existence of pleomorphic adenoma and Actinomycosis in the infratemporal fossa.

Case description: A 40-year-old female patient complains of pain over the upper left back tooth region for the past 6months. The patient had a past history of a small lesion in 28 region which resulted in constant irritation. The patient had consulted the dentist for the same and underwent extraction of 28 and 38 three years back, which was thought to be the cause of the swelling.

Conclusion: The case highlights the importance of proper history taking, clinical examination, and histopathological examination in arriving at a precise diagnosis of a lesion irrespective of size and clinical presentation.

Keywords: Pleomorphic adenoma, actinomycosis, infratemporal fossa, mixed tumor, salivary gland tumors.

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Introduction

Salivary gland tumors are the rare entity of head and neck regions with the majority being benign and 20% being malignant. Pleomorphic adenoma is the most common benign neoplasm that accounts for about 70% of all salivary gland tumors. The rare sites include the throat (2.5%), retromolar region (0.7%), the floor of the mouth, and the alveolar mucosa. It exhibits a vast morphological assortment with a mixture of ductal and myoepithelial elements along with significant variability in the mesenchymal components. Hence the name mixed benign tumor.[1]

Actinomycosis is a bacterial infection, the causative organism being a gram-positive anaerobic bacterium *Actinomyces israelii*. The bacteria usually affect the four regions namely cervicofacial, thoracic, abdominal, and genital. Among these, the most common site is the cervicofacial region. The casein the cervicofacial region presents as a draining sinus near the mandible. The suppurative reaction of the infection discharges a large yellowish fleck that represents colonies of the bacteria called sulfur granules.[2]

In this case report, we are discussing a rare case of pleomorphic adenoma co-existing with actinomycosis of an unusual site i.e., the infratemporal fossa.

Case Report

A 40-year-old female patient complains of pain over the upper left back tooth region for the past 6months. The patient had a past history of a small lesion in 28 region which resulted in constant irritation. The patient had consulted the dentist for the same and underwent extraction of 28 and 38 (both upper and lower 3rd molar on left side) three years back, which was thought to be the cause of the swelling. The patient was relatively symptomless after the extraction. The patient reported pain in the same region one and half years ago, which was continuous in nature and of pricking type. As there was a progression in pain, the patient was advised to perform Cone Beam Computed

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Tomography by an Otorhinolaryngologist. The patient was then referred to the department of oral and maxillofacial surgery of our institution.

On extra-oral examination, a mild facial asymmetry was noted on the left side of the face with a diffuse swelling over the left lower one-third region. The swelling was diffuse, soft in consistency, and tender on palpation. Submandibular lymph nodes were also palpable and tender.

On intra-oral examination, a diffuse swelling obliterating the left buccal vestibule extending mesiodistally from 26 to 28 region was observed (Fig.1). The mucosa over the swelling appeared erythematous with a sinus opening and pus discharge in 26 region. The swelling was soft in consistency, non-fluctuant and tender on palpation.

CBCT examination reveals a well-defined, non-enhancing soft tissue lesion with a smooth margin in the left infratemporal fossa measuring about 2.8 x 1.7 x 3.5cm in diameter. The lesion also resulted in thinning and focal erosion of the posterior wall of the

Table 1: Review of literature of pleomorphic adenoma at infratemporal fossa

Author	Year	Patient age	Gender	Site	Size of the lesion
Jeyanthi K *et al*[6]	2007	45years	Female	Right infratemporal fossa	3.1 x 2.2 cm in diameter
Hadi El *et al*[7]	2009	59years	Female	Right infratemporal fossa	3.3 X 1.8 cm in diameter
Gurey LE *et al*[8]	2010	42years	Male	Left infratemporal fossa	-
Wong DK *et al*[9]	2019	61years	Female	Metastatic PA in Right infratemporal fossa	2.5 x 2.5 cm in diameter
Present case report	2021	40years	Female	Left infratemporal fossa	2.8 x 1.7 x 3.5cm in diameter

Table 2: Review of literature of actinomycosis at infratemporal fossa

Author	Year	Patient age	Gender	Site	Size of the lesion
Carrau RL *et al*[11]	1993	6years	Male	Right Infratemporal fossa	3.5 x 4 cm in diameter
Sethi A *et al*[10]	2008	43years	Female	Left Infratemporal fossa	-
Kinard BE *et al*[12]	2016	50years	Male	Left infratemporal fossa	-
Hotte GJ *et al*[13]	2019	58years	Male	Left infratemporal fossa – extension from maxillary lesion	-

Fig. 1: Intraoral Findings- A small lesion on the left maxillary tuberosity region.

Fig. 2: Cone Beam Computed Tomography - Radio-dense area in the left infratemporal region.

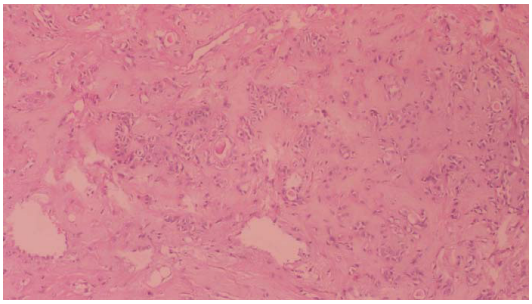

Fig. 3: Epithelial and myoepithelial cells in the form of cords, sheets and duct like pattern (H and E, 10x).

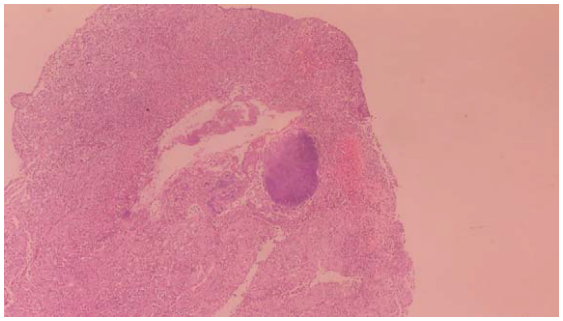

Fig. 4: Connective tissue stroma with hyalinized areas and actinomycotic colony (H and E, 4x).

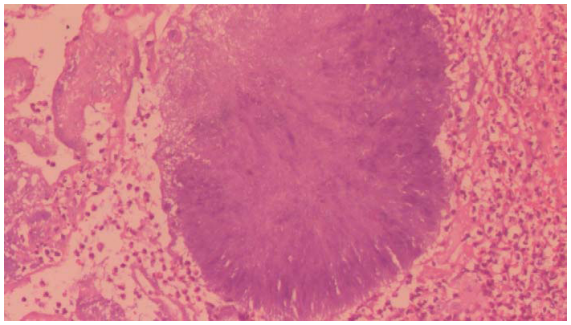

Fig. 5: Actinomycotic colonies (H and E, 20x)

left maxillary sinus (Fig.2).

Based on the clinical and the radiographic findings, a provisional diagnosis of odontogenic tumor was given. Accordingly, an incisional biopsy was taken and the tissue was subjected to histopathological examination.

Histopathological examination revealed a biphasic lesion comprising of epithelial and myoepithelial cells arranged in cords, sheets, and duct-like patterns (Fig.3). The cells are round to oval, having a bland nucleus, inconspicuous nucleoli, and scant to moderate cytoplasm. The connective tissue stroma showed abundant loose chondromyxoid and hyaline matrix (Fig.4). The section also showed few actinomycotic colonies surrounded by chronic inflammatory cells (Fig.5).

Histopathological findings confirmed the diagnosis of Pleomorphic adenoma co-existing with actinomycosis. The lesion was surgically excised under general anesthesia through an intra-oral approach with proper debridement. The patient was relatively symptom free.

Discussion

The term "pleomorphic adenoma" was coined by Willis in 1953, attributing to its distinct and diverse mixed histopathology.[3] It makes up the largest proportion of all the benign salivary gland neoplasm but it rarely involves the minor salivary glands which account for about 6.4%. The rare locations of pleomorphic adenoma reported are in the scalp, eyelid, nose, cheek, upper lip, and external auditory canal.[4]

The lesion occurring in the infratemporal region is generally rare because of its anatomic location. The cases appear clinically as a mass obliterating the buccal vestibule. Similarly, our case was also clinically present as a mass obliterating the left buccal vestibule.[5] The most common lesions occurring in the infratemporal fossa are nasopharyngeal carcinoma, juvenile angiofibroma, and adenoid cystic carcinoma. Pleomorphic adenoma arising in the infratemporal region is a rare possibility. (Table.1).

Actinomycosis is an infection caused by gram-positive, microaerophilic, facultative anaerobic, and non-acid fast branched filamentous bacteria called *Actinomyces israelii*. The organism colonizes the oral cavity and oropharynx and causes cervico-facial actinomycosis by entering the subcutaneous tissue or tonsillar fossa due to trauma.[10] The other forms of the disease are pulmonary actinomycosis and abdominal actinomycosis. The cervico-facial actinomycosis is the most commest form and the lesion presents as an indolent, insidiously growing mass with surrounding induration in the mandible. If left untreated, the lesion presents as multiple discharging sinuses or as a draining abscess.[11]

The other unusual sites of actinomycosis so far reported are malar and cranial bone, trigeminal ganglion, infraorbital nerve, parotid gland, tongue, and posterior pharyngeal wall.[11] There are only a few actinomycosis cases reported in the infratemporal fossa in the literature (Table.2).

In this present case, it can be attributed that the patient had the pleomorphic adenoma 3 years back itself, when she complained of a ball-like swelling in the tuberosity region. However, this was ignored by a general dentist and considered it as a reactive lesion because of impingement from the lower third molar and suggested extraction of both upper and lower third molars. The lesion probably has further grown in size from the tuberosity region because of long duration and along with the superadded Actinomycotic infection, the lesion has infiltrated into the infratemporal fossa.

Conclusion

In conclusion, we attempt to highlight the presence of Actinomycotic infection which is extremely rare co-existing with pleomorphic adenoma. The presence of the infection has weakened the bone and influenced the extension of lesion into the infratemporal fossa, acting as an adjunct to behave like an aggressive lesion. This case also highlights the importance of proper history taking, clinical examination, and histopathological examination in arriving at a precise diagnosis of a lesion irrespective of size and clinical presentation. In the English literature, there is no such case report of pleomorphic adenoma of infratemporal fossa coexisting with Actinomycosis.

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