

# PLEOMORPHIC ADENOMA IN THE SUBMANDIBULAR GLAND: A CASE REPORT

**Authors:** Grace Budhiraja (1) \*, Navjot Kaur (2) Harsimrat Singh (3)

**Authors Affiliations:** (1) Professor, (2) Assistant Professor, (3) Senior resident, Department of ENT, Adesh institute of medical sciences and research , Bathinda, Punjab

## ABSTRACT

Neoplasms of salivary glands represent a small group among the diseases involving the head and neck complex. In this group, the pleomorphic adenoma is the most frequent neoplasm, yet involves the submandibular gland in only 12.8 % of cases. A patient presenting a swelling in the region of the submandibular gland was admitted for submandibulectomy and sent for histopathological examination. All specimens were histologically defined as pleomorphic adenoma. This multicentric finding is of great interest, perhaps explaining the recurrence rate of this neoplasm. The patient is in continuous follow-up and has not presented signs of recurrence.

## KEYWORDS

Pleomorphic adenoma, Submandibular gland, Salivary gland tumor, Benign neoplasm, Multicentric tumor

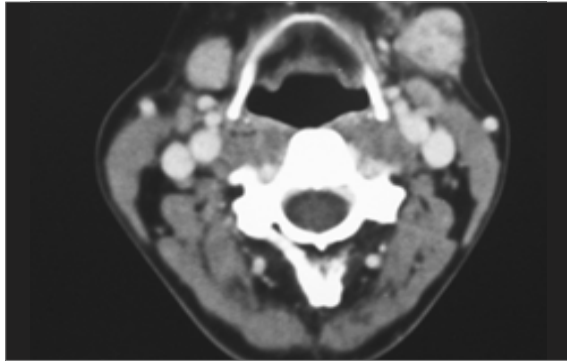
## INTRODUCTION

Neoplasms of salivary glands represent a significant group of neoplasms that involves the head and neck complex. In this group, the pleomorphic adenoma is the most frequent neoplasm, yet the submandibular gland is involved in only 12.8 % of cases. A rare occurrence is its multiple manifestation within the gland, around 2.6%, presenting as multicentric or multifocal. Besides the rarity, this variant represents a determinant characteristic for the recurrence potential, either because of the greater number of lesions or due to its invasive nature [1-5]. This paper reports a case of a

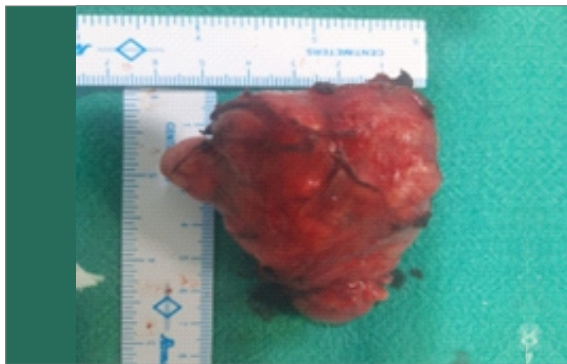
pleomorphic adenoma originating from the submandibular gland, in which surgical interventions demonstrated a multicentric behavior of the lesion.

## CASE REPORT

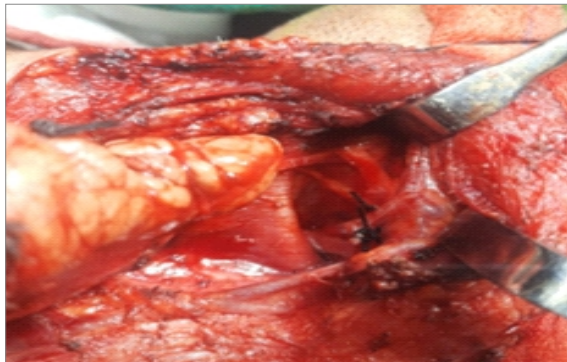
A male patient, 37-years-old, presented with a painless swelling in the region of the submandibular gland, reporting 18 months of evolution. Clinical examination disclosed an endophytic nodule about 3.5 cm, localized in the region of the left submandibular gland, with lobulated surface and firm consistency. Computed tomography scan demonstrated an increase in the size of the submandibular gland without change of its density, suggesting a neoplasm (Fig.1). A fine needle aspiration cytology was done , give impression of pleomorphic adenoma. The result of the histopathologic examination, in samples, was pleomorphic adenoma. Interestingly, during the surgical procedure two additional nodules were observed just beside the gland, and were removed (Fig.2). After the identification of the gland capsule, a subcapsular enucleation was accomplished, avoiding injury to the lingual and hypoglossal nerves at the medial aspect of the gland (Fig.3). Total removal was done with identification and clamping of the salivary duct at its most anterior portion. All specimens were analyzed and histologically defined as pleomorphic adenoma. The associations of these findings characterized the multicentric behavior of the disease. The patient is being followed and , presents with good mobility and sensibility of the tongue. No signs of recurrence were found.



**Figure 1:** Scan showing neoplasm



**Figure 2:** Delivered specimen



**Figure 3:** Gland with lingual and hypoglossal nerves

## DISCUSSION

The term pleomorphic adenoma was initially suggested due to the diversity of histological patterns observed in these tumors, basically characterized by ductal and myoepithelial neoplastic components, immersed in a hyalinized collagenous stroma with incomplete capsular involvement in most cases. This tumor presents itself as a painless and slow growing nodule that

can reach great dimensions without compromising the adjacent anatomical structures [1-5]. Proper treatment is only achieved when the correct diagnose is obtained. Fine needle aspiration is the preferred initial diagnostic procedure since incisional biopsy for salivary gland lesions may result in seeding tumor beyond the capsule. However, due to the lack of a typical histological pattern and to the morphological variety of myoepithelial neoplasms, fine needle aspiration biopsy, incisional biopsy or frozen biopsy are useful in the differentiation from malignant lesions, but do not always provide for a definitive diagnosis. This confirmation is only obtained from the histopathologic examination of the entire specimen, associated with clinical and image information [4-7]. Treatment success is directly related with total removal of the gland. Once the pleomorphic adenoma presents an incomplete capsule, its enucleation with the preservation of the gland is controversial, probably increasing the possibilities of recurrence [8,2]. Recurrences generally present as multiple nodes in the topography of the affected gland. This characteristic, however, may not be exclusive to recurrences, being seen in primary lesions as well [2-9]. Multiple nodes affecting the same region characterize the multicentric or multifocal behavior of a disease. This characteristic is rare and mostly seen in malignant neoplasms, being interpreted, somehow, as local metastasis [10]. Multicentricity will increase the possibility of recurrences, since an invasive characteristic is not supported in the literature [10-12]. In the report of 38 cases of submandibular pleomorphic adenoma presented by Laskawi et al., the only multicentric case reported developed a recurrence [8]. Another variation of the behavior of a lesion is the possibility of its presentation in different locations at the same time, being then called as a synchronous lesion, mostly seen in Whartin tumor [12]. Seeding of the tumor during surgical procedures has been the primary feature that explains multicentric behavior; however, in this case report, multiple presentations were already seen in the incisional biopsy. Another detail is that

the submandibular gland maintained capsular integrity; therefore the possibility of local metastasis would be highly questionable. Epithelial remnants of the submandibular gland trapped in local lymph nodes could also explain multicentric and synchronous behavior of the presented case [12]. Due to the reports of this variant behavior of a pleomorphic adenoma, the authors support the importance of removal of nodes adjacent to the affected gland, even though they seem to be lymph nodes, and they should all be histologically analyzed. This would probably lead to an additional few minutes of surgery, but this approach can avoid secondary surgery.[2,8,10,11].

## CONCLUSION

Knowledge of the clinical behavior of a neoplasm is important to promote adequate treatment. Pleomorphic adenomas have a tendency to reoccur but secondary surgery for recurrent lesions carries a worse prognosis.

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**\*Corresponding Author-**

Dr. Grace Budhiraja  
Professor, Department of ENT  
Adesh institute of medical sciences  
drgracebudhirajaadeshent@gmail.com  
9464780122

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