

Case Report

Pleomorphic adenoma with lipometaplasia: Rare histology finding of a common entity

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Article history: Received 14-10-2023 Accepted 10-11-2023 Available online 11-12-2023	We report a rare case of a pleomorphic adenoma with lipometaplasia. A 32-year-old woman presented with intraoral swelling over right side of palate. Computed tomography revealed a heterogeneously enhancing right hard palatal mass without bony erosions. Preoperative diagnosis was offered with intraoral fine needle aspiration cytology following which excision was performed. On histopatholgy, final diagnosis of pleomorphic adenoma with lipometaplasia offered. Though rare it must be kept in mind as differential in cases of fat containing salivary gland lesions.
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1. Introduction

Pleomorphic adenoma is the commonest salivary gland benign neoplasm with majority of cases occurring in parotid and less commonly in minor salivary glands. These mixed tumours show proliferation of both epithelial / myoepithelial cells and chondromyxoid stroma. Metaplastic changes are common in both epithelial as well as stromal components. These include Squamous, columnar, sebaceous, oncocytic, mucinous, myxoid, hyaline, cartilaginous and osseous differentiations. However, lipometaplasia with stromal adipocytic component is quite rare.^{1,2}

2. Case Presentation

A 32-year-old female presented with complaints of intraoral swelling since last three & half months with associated difficulty in swallowing food. She denied any significant past or family history. Intraoral examination showed a 4x3 cm sized globular exophytic lesion over right side of

hard palate which was smooth in appearance, non tender and firm on palpation. However exact borders were not

Intraoral fine needle aspiration cytology (FNAC) attempted from the lesion, showed moderately cellular smears having classical morphology with admixture of epithelial / myoepithelial & characteristic magenta coloured fibrillary chondromyxoid mesenchymal element.

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traceable. Extraoral examination was within normal limits. As a part of investigations, intraoral ultrasonography (USG) & computed tomography (CT) maxilla plain & contrast studies carried out. USG showed a well-defined lesion with multiple dense internal echoes & septations within giving posterior acoustic enhancement with minimal vascularity on colour doppler. While CT Maxilla Plain & contrast showed ill-defined heterogeneously enhancing soft tissue mass density lesion posterolaterally reaching upto retromolar trigone while superiorly upto floor of maxillary sinus and posteriorly involving soft palate leading to asymmetry of pharyngeal lumen (CT attenuation value of 60-70 HU). No bony erosions seen. Both imaging studies favoured benign lesion.

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Background showed scanty foamy macrophages & anucleate squams. Hence, diagnosis of pleomorphic adenoma (PA) offered. Excision of intraoral swelling was done which showed a well circumscribed partially encapsulated nodular lesion measuring 2.5x1.5x1 cm. Both external & cut surface were homogenous greyish white in appearance. Histopathological examination showed partially encapsulated lesion with admixture of epithelial and mesenchymal components. Mesenchymal component was noted in the form of chondromyxoid material along with around 30% of areas showing mature adipose tissue fragments. Mesenchymal elements were intricately mixed with epithelial elements including both acinar & myoepithelial cells. Squamous keratin pearls were also seen. There was no atypia, perineural or lymphovascular invasion noted. Based on overall findings diagnosis of pleomorphic adneoma with lipometaplasia offered. (Figure 1) Unfortunately patient lost to follow up.



Figure 1: a): Intraoral right hard palatal swelling with smooth external appearance; b): FNAC picture showing epithelial/ myoepithelial & chondromyxoid mesenchymal component admixture (May Grundwald Giemsa stain, 40x), c): Scanner view of PA with epithelial/myoepithelial & chondromyxoid mesenchymal component admixture with matureadipose tissue (Haematoxylene and Eosin stain) d): Lipomatous component (Haematoxylene and Eosin stain, 40x)

3. Discussion

Ng et al. (1995) first reported case of PA with extensive lipometaplasia in submandibular glands.^{2,3} Seifert et al. defined lipomatous PA as pleomorphic adenoma with more than 90% lipomatous component of total tumour.⁴ Haskel et al. suggested myoepithelial origin for lipometaplasia as they have ability to transform into various metaplasia.⁵ To the best of our knowledge, 20 cases of lipomatous PA / PA with lipometaplasia of salivary glands are reported till

date among which 2 were reported in locations like trachea⁶ and parapharyngeal space.⁷ Few lipomatous mixed tumour cases are also reported in locations like scalp skin, breast and ceruminous glands of external auditory canal.²

Most of the cases are seen in young age having age range of 14 to 74 yrs with slight female preponderance. Most of cases are located in parotid followed by minor salivary glands of hard palate and submandibular glands (single case). PA with lipometaplasia cases are usually well circumscribed ranging from 1 to 7 cm in maximum dimension with largest noted in a parapharyngeal space location.⁷ Lipomatous component ranged from 25% to more than 95% of tumour volume. Preoperative USG & CT scan was done in most of the cases with few scans suggesting benign lesion with high fat contents. Preoperative FNAC diagnosis of PA with lipometaplasia was offered in two cases.² Most of cases showed no recurrence on follow up. However, one case was identified in a recurrent growth with absence of lipomatous component in primary lesion. Singh et al & Brisebois et al have reported PA with concurrent extensive squamous metaplasia and adipocytic metaplasia.8

Overall fat containing salivary gland neoplasms are rare. Agaimy et al⁹ have classified fat containing salivary gland lesions into four broad categories including fat containing epithelial / myoepithelial tumours, mixed lipoepithelial tumours, true adipose tissue neoplasms and tumour like fatty lesions. Among these, lipoma (soft tissue origin), oncocytic lipoadenoma, sialolipoma, and pleomorphic adenoma/myoepithelioma with lipometaplasia are common entities in decreasing order of frequency. Lipoma variants, atypical lipomatous tumour / liposarcoma (soft tissue) and uncommon polycystic lesions like striated duct adenoma, lipoadenoma with probable striated duct differentiation, parotid lipoadenoma with sclerotic & polycystic changes and microcystic lipoadenoma tumours are quite rarely reported in salivary glands. Tumour like fatty lesions like diffuse / interstitial lipomatosis or lobular fatty atrophy can be mimicker in a small biopsy or FNAC samples.⁹

4. Conclusion

Thus, though rare, pleomorphic adenoma with lipometaplasia should be kept in mind while assessing fat containing salivary gland lesions especially in fine needle aspiration & small biopsy specimens.

5. Source of Funding

None.

6. Conflict of Interest

None.

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