

Parapharyngeal space pleomorphic adenoma with extensive lipometaplasia – A rare unique pathological entity

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Abstract

Pleomorphic adenoma (PA) is the most common neoplasm of the salivary gland. Fat-containing tumors encountered in salivary glands are very rare. We had a case of PA of parapharyngeal space and surprisingly the final histopathology showed an unusual finding of lipometaplasia. We believe that this is the first case from parapharyngeal space to be reported in the literature. The aim of this report is to discuss the behavior of lipometaplasia in PA with review of literature. Pathological characterization of this rare tumor might help in proper classification and enhance their recognition.

Keywords: Lipometaplasia, parapharyngeal space, pleomorphic adenoma

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Submitted: 25-Mar-2020, **Revised:** 18-Jan-2021, **Accepted:** 19-Jan-2021, **Published:** 19-Mar-2021

INTRODUCTION

Primary parapharyngeal space tumors are very rare, accounting for only 0.5% of neoplasms of head and neck. Most of these tumors (70%–80%) are benign and 40%–50% of these tumors originate in the minor salivary glands, specifically pleomorphic adenoma (PA).^[1,2]

The classical appearance of PA is characterized by a mixture of myoepithelial and ductal cells, with foci of chondromyxoid areas admixed with areas of squamous and/or sebaceous metaplasia. Occasionally, areas of lamellar bone with osteoblasts and osteoclasts representing osseous metaplasia and focal collections of mature adipocytes, which may be due to entrapment of surrounding fibroadipose tissue, can be present. Although scattered, isolated adipocytes might be rarely encountered in classical PAs. PAs and myoepitheliomas with extensive

lipometaplasia comprising more than 20% of the tumor is exceedingly uncommon with about 15 reported cases in the English literature to date.^[3]

Most of the cases are reported from major salivary gland especially from parotid gland and few cases from minor salivary glands of oral cavity. Our case is the first reported case from parapharyngeal space. The clinical significance of this unusual finding of lipometaplasia is not clear. Our aim of this report is to discuss the behavior of lipometaplasia in PA with review of literature.

CASE REPORT

A 34-year-old man with no comorbidities, presented with the difficulty in opening mouth with intermittent pain in the right ear for the past 1 year. On examination of oropharynx,

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How to cite this article: Mani S, Anto R, Michael RC, Ponmar M. Parapharyngeal space pleomorphic adenoma with extensive lipometaplasia – A rare unique pathological entity. *J Oral Maxillofac Pathol* 2021;25:S4-6.

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10.4103/jomfp.JOMFP_117_20

mucosa covered soft bulge was present in the soft palate on the right side and also pushing the right tonsil medially. Neck examination was found to be normal. Magnetic resonance imaging (MRI) face [Figure 1] was done which revealed a well-defined encapsulated heterogeneously enhancing mass in the right prestyloid parapharyngeal space measuring 62 mm × 37 mm × 61 mm, with no infiltration to adjacent structures or skull base extension. These features are in favor of mass likely originating in the minor salivary gland. Thus, with the preoperative diagnosis of a benign right parapharyngeal space tumor, transcervical excision of tumor was done. A well-defined 7 cm × 6 cm × 4 cm mass was visualized in the right prestyloid space intraoperatively and was excised completely. Intraoperative and postoperative period was uneventful.

Microscopically, sections show parts of a circumscribed tumor composed of an intimate admixture of epithelial, myoepithelial, and stromal components. The epithelial component is composed of anastomosing strands, cords, trabeculae, and tubules with mildly pleomorphic nuclei, fine chromatin, few with visible nucleoli, and moderate eosinophilic cytoplasm surrounded by an outer mantle of myoepithelial cells. The plasmacytoid to spindled myoepithelial cells with moderate amounts of eosinophilic to clear cytoplasm are also arranged in small clusters and cords, which blends into the surrounding chondromyxoid stroma. There is focal oncocytic and squamous metaplasia. There is extensive adipocytic metaplasia constituting more than 90% of the tumor volume. There is no necrosis. There is no evidence of malignancy. Thus, with the following above features, the diagnosis of PA with extensive lipometaplasia was made [Figure 2]. The patient was followed up to 6 months and no recurrence was noted.

DISCUSSION

PA with extensive lipometaplasia is a recently described histologic variant of PA that demonstrates varying degrees

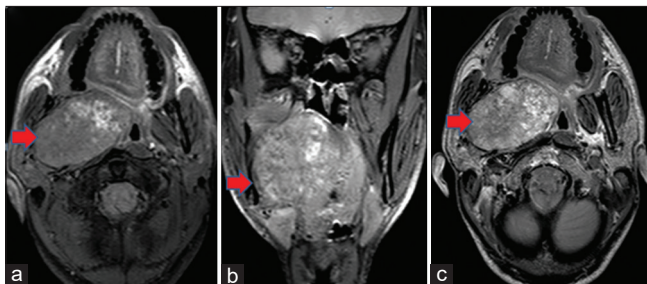


Figure 1: MRI face showing tumor in right parapharyngeal space (red arrow) (a) T1 W axial image showing multiple punctuate hyperintensities within the tumor. (b) T1W coronal image showing same findings as axial image. (c) T1W GADO contrast showing heterogeneous post contrast enhancement

of lipocytic differentiation.^[4] Haskell *et al.* suggested that it could be due to the ability of myoepithelial cells to undergo various metaplasias is the cause of the unusual histologic appearances of this tumor. The adipocytic components comprised 20%–90% of the tumor mass in most cases.^[5]

Due to their rarity, there is no general agreement on the histogenetic classification and nomenclature of these lesions and a few lesions are not included in the current World Health Organization Classification of head and neck tumors. Agaimy *et al.*, described a classification system for fat containing tumors and tumor-like lesions of salivary glands which can be divided into monophasic true adipocytic neoplasms (lipoma and its variants and atypical lipomatous tumor/liposarcoma) and hybrid lipoepithelial lesions composed of epithelial derivatives admixed with a variable fatty component (PA and myoepithelioma). In addition, rare salivary gland tumors may display a fatty component associated with an epithelial component distinct from the above entities (sialolipomas and lipoadenomas).^[3,6] In the present case, the presence of epithelial and myoepithelial in a background of chondromyxoid stroma, with the presence of 90% of lipometaplasia points to the diagnosis of PA with extensive lipometaplasia.

Haskell *et al.* in his study, reported that approximately 80% of PA involved major salivary glands (11/12 in the parotid) with only three cases in minor salivary glands of oral cavity.^[5] In our case, the tumor is from parapharyngeal space which has not been previously reported in literature. None of the reported case in the literature had recurrence after surgical excision.^[7-12] PA with lipometaplasia behaves in the same way as their nonfat-containing counterparts; both types can be cured with simple excision and do not carry a risk of recurrence.^[12]

MRI Imaging can predict the fat component of the tumor but the final characterization will be totally depends on histopathological features.^[8] Most of the time, the diagnosis of lipometaplasia will be made after the curative resection of the tumor which does not have much clinical

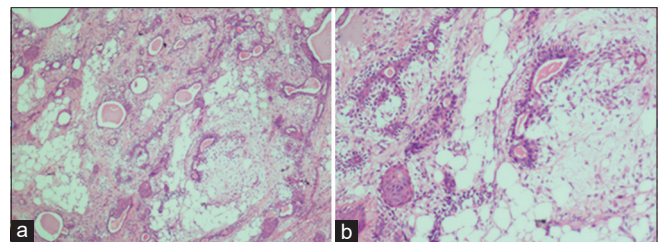


Figure 2: Hematoxylin and Eosin stain shows all components—tubules lined by epithelial and myoepithelial cells; chondromyxoid stroma and the lobules of fat (a) 40X view. (b) 100X view

significance but pathological characterization might help in proper classification of this rare tumor and enhance their recognition. Thus, might help us to clarify their pathogenesis and biological behavior of the tumor. Even though an uncommon histopathological variant, it is treated like as common variant because of the rarity and benign in nature.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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