Case Report

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20204916

Sclerosing variant of mucoepidermoid carcinoma: a case report and review of literature

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Received: 30 August 2020 Accepted: 03 October 2020

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ABSTRACT

Mucoepidermoid carcinoma, a malignant tumour of the salivary gland is histologically characterized by the presence of mucoid cells, epidermoid cells and intermediate cells. There are many variants of this tumour. A particular variant, sclerosing variant of mucoepidermoid carcinoma is presented due to its rarity and chances of misdiagnosis as benign lesions due to the presence of extensive sclerosing stroma.

Keywords: Mucoepidermoid, Salivary gland, Sclerosing, Intermediate

INTRODUCTION

The most common malignant tumour of the major salivary gland is mucoepidermoid carcinoma, which is histologically characterized by the presence of mucoid cells, epidermoid cells and intermediate cells.¹ The variants include oncocytic, sclerosing, unicystic, clear cell, goblet cell, spindle cell, psammomatous and sebaceous types.²

High grade mucoepidermoid carcinoma is characterized by the presence of more solid areas, compared to cystic areas, which are prominent in low grade.³ Sclerosing variant of mucoepidermoid carcinoma is characterized by extensive sclerosis and paucity of cells, which can lead to misdiagnosis.⁴

Here, we report an interesting case of sclerosing variant of mucoepidermoid carcinoma.

CASE REPORT

A 36 year old female presented with history of gradually increasing painless swelling in the left parotid region since

6 years. Clinically, there was a 3×2 cm firm to hard swelling in front of ear in the left parotid region. Fine needle aspiration cytology (FNAC) performed was acellular. Left parotidectomy was done.

Gross examination revealed a circumscribed tumour measuring $3 \times 2.5 \times 2.5$ cm with a firm to hard cut surface. Cut section was uniform tan, firm to hard with few glistening areas.

Microscopic examination revealed parts of a well circumscribed tumour with extensive stromal sclerosis. There were few nests and small cysts lined by mucin producing columnar cells and intermediate cells (Figure 1 and 2).

Occasional cystic spaces were filled with mucoid material. This cellular component comprised only about 10% of the tumor. There was no squamous cell component.

There was no lymphovascular or perineural invasion. Biopsy was reported as sclerosing variant of low grade mucoepidermoid carcinoma, Brandwein grade 2.

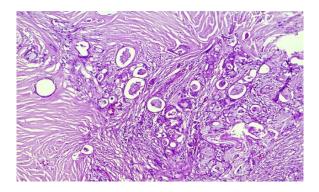


Figure 1: Extensively sclerotic stroma with nests and small cysts of tumour cells, hematoxylin and eosin (H and E) 100x.

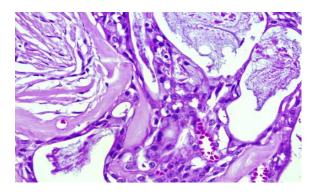


Figure 2: Mucinous cells and intermediate cells, (H and E) 400x.

DISCUSSION

Tumours of the salivary gland can be benign or malignant and can occur in major and minor salivary glands. Pleomorphic adenoma is the most common benign tumour and mucoepidermoid carcinoma is the most common malignant tumour in the parotid gland.⁵ Mucoepidermoid carcinoma constitutes 10% of salivary gland neoplasm and 35% of malignant neoplasms of the salivary gland.⁶ Mucoepidermoid carcinoma is composed of mucin producing cells, squamous cells, intermediate cells and clear cells.⁷ Sclerosing variant of mucoepidermoid carcinoma is characterized by extensive sclerosis and paucity of cells, which can lead to misdiagnosis.⁴

The behavior of mucoepidermoid carcinoma is strongly correlated with the clinical stage and histologic grade.⁸

First case of mucoepidermoid carcinoma was reported by Chan and Sawin 1987.⁹

Size of the tumour (more than 2 cm) is one of the most important prognostic factor in sclerosing mucoepidermoid carcinoma according to Fadare et al.¹⁰ Those cases in which tumour size was more than 2 cm had metastasis.

The differential diagnosis of sclerosing mucoepidermoid carcinoma includes necrotizing sialometaplasia, mixed tumors, sclerosing polycystic adenosis, hyalinizing clear cell carcinoma, sclerosing sialadenitis and polymorphous low-grade adenocarcinoma.¹¹

The lobular architecture is maintained in case of necrotizing sialometaplasia. An important diagnostic clue to the diagnosis of sclerosing mucoepidermoid carcinoma is the presence of keloid like sclerosis and peripheral rim of lymphocytes. In the other benign conditions, lobular architecture is maintained. Few clues to distinguish from other conditions includes the following.¹²

A distinct feature which helps in distinguishing sclerosing mucoepidermoid carcinoma from sclerosing polycystic adenosis is the presence of bright eosinophilic granules in some cells in sclerosing polycystic adenosis.

Features helping in distinguishing sclerosing mucoepidermoid carcinoma from pleomorphic adenoma with squamous and mucinous metaplasia includes lack of invasive growth and presence of plasmacytoid hyaline cells and chondromyxoid stroma. Another important point to be noted in low grade mucoepidermoid carcinoma is never frankly squamous in appearance nor shows evidence of keratinisation.

Chronic sclerosing sialadenitis in most cases is considered to be a manifestation of immunoglobulin G4 (IgG4) related disease and almost exclusively affects submandibular gland. IgG4+ plasma cell count >100/hpfand IgG4/IgG ratio >40% can help in the diagnosis.

Polymorphous low-grade adenocarcinoma has an infiltrative growth and almost exclusively occurs in minor salivary glands.

Hyalinising clear cell carcinoma shows presence of clear cells and cells with eosinophilic cytoplasm in a hyalinised and myxoid stroma. Molecular analysis for t (12:22) (q13; q12) EWSR1-ATF1 can help in confirming the diagnosis of hyalinising clear cell carcinoma.

FNAC is most often inadequate due to the abundant sclerotic stroma.

CONCLUSION

This case of sclerosing mucoepidermoid is presented to highlight the importance of differentiating it from the other benign mimickers in the salivary gland.

ACKNOWLEDGEMENTS

Authors would like to extend their sincere thanks to Dr. Sheila Nair, HOD, Department of Pathology, Sri Narayani Hospital and Research centre for helping with the diagnosis of this particular case.

Funding: No funding sources Conflict of interest: None declared Ethical approval: Not required

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Cite this article as: Radhika S, Burad D, Sonwane TY, Viswanath B. Sclerosing variant of mucoepidermoid carcinoma: a case report and review of literature. Int J Res Med Sci 2020;8:4124-6.