

Polymorphous Low Grade Adenocarcinoma: A Report of Two Cases

¹Sumit Majumdar, ²Jay Gopal Roy, ³Divya Uppala

ABSTRACT

Polymorphous low grade adenocarcinoma (PLGA) is a difficult lesion to diagnose both clinically and histopathologically due to its varied presentation and its morphological diversity. Polymorphous low grade adenocarcinoma is a relatively new entity which arises from the salivary glands. We are reporting two cases PLGA with an unusual histopathological picture.

Keywords: Polymorphous, Low grade, Adenocarcinoma, Report, Oral, Histopathology.

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INTRODUCTION

Polymorphous low grade adenocarcinoma (PLGA) was described as a distinct entity by Freedman and Lumerman in 1983. This lesion was previously misdiagnosed as pleomorphic adenoma, monomorphic adenomas, malignant pleomorphic adenomas and adenoid cystic carcinomas.^{1,2} Evans and Batsakis coined the term 'PLGA' in 1984.³ It is a fairly uncommon lesion and, if it occurs, its usual site is the minor salivary gland.^{4,5} In this article, we present a review of literature and case reports of two cases encountered in Dr R Ahmed Dental College and Hospital, Kolkata.

CASE REPORTS

Case 1

In the outpatient Department of Oral and Maxillofacial Pathology of Dr R Ahmed Dental College and Hospital, a 60-year-old female patient presented with a slowly enlarging asymptomatic growth involving the right mid facial region

for the last 9 years (Fig. 1). Radiological investigation revealed diffuse radiopacity involving the entire right side of the mid face. Computed tomography (CT) of middle one third of face revealed a large space occupying lesion with involvement of the maxillary antrum and destruction of all the bony boundaries except the orbital floor. Intraoral examination revealed the presence of a large, firm asymptomatic growth involving the right posterolateral aspect of palate. The palatal mucosa was ulcerated and regional teeth were slightly mobile. Section stained with hematoxylin and eosin revealed the presence of proliferating neoplastic glandular patterns having prominent bland nuclei with minimal cytoplasm. Interestingly, multiple areas of lobular arrangement with conspicuous palisaded peripheral cells were also noted in the same section (Figs 2A and B).

Case 2

A male patient, 66 years of age reported with a large slowly enlarging painful swelling involving the right side of the face for the last 10 years (Fig. 3), with a history of previous surgical intervention (hemimandibulectomy) done about 10 years back for a swelling in the submandibular region. The overlying skin was normal but showed prominent vascular marking. Radiological presence of 'K' wire in the right side of the mandible replacing the resected part. Computed tomography of lower 1/3rd revealed a large space occupying lesion. Intraoral examination revealed a large, soft, lobulated nonulcerated growth involving the entire buccal mucosa, buccal sulcus and the floor of the mouth

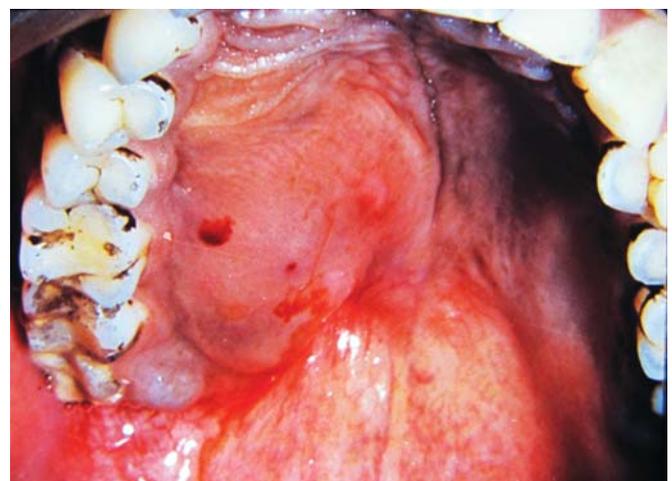


Fig. 1: Growth involving the right midfacial region

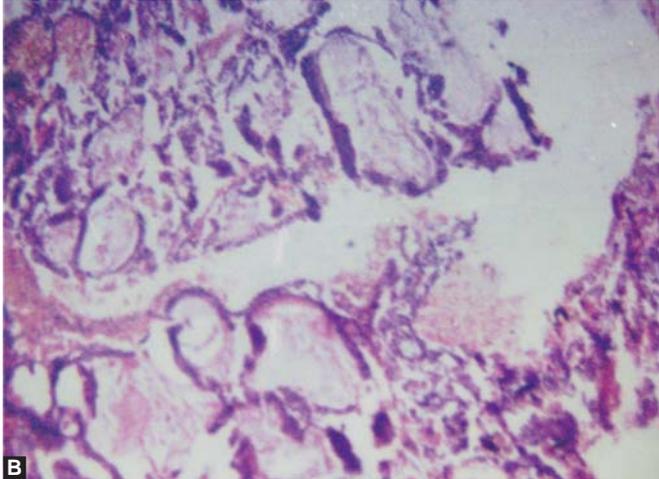
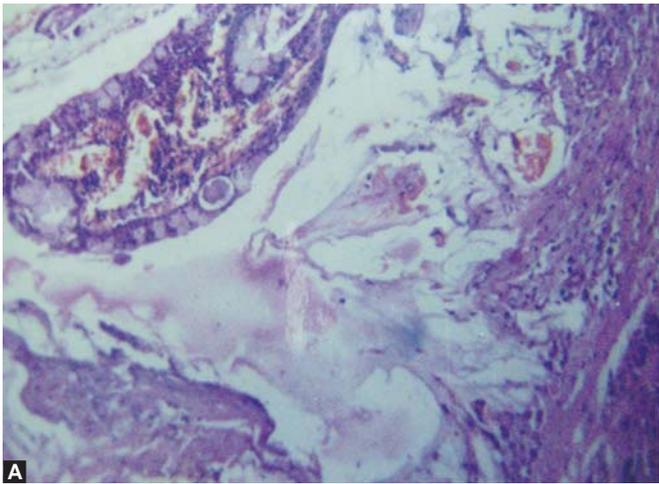
^{1,2}Head, ³Assistant Professor

¹Department of Oral Pathology, Gitam Dental College Visakhapatnam, Andhra Pradesh, India

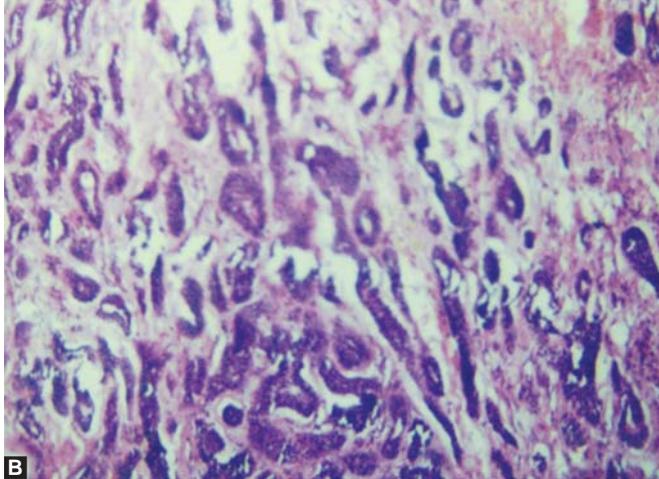
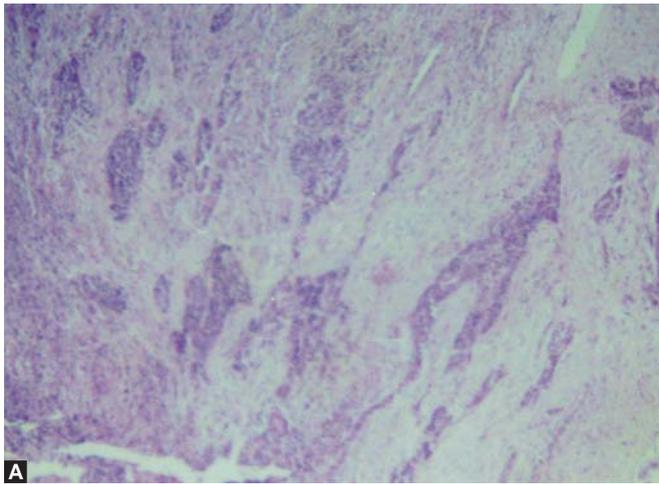
²Department of Oral Pathology, Dr R Ahmed Dental College and Hospital, Kolkata, West Bengal, India

³Department of Oral and Maxillofacial Pathology, Gitam Dental College, Visakhapatnam, Andhra Pradesh, India

Corresponding Author: Divya Uppala, Assistant Professor Gitam Dental College, Visakhapatnam-530 045, Andhra Pradesh India, Phone: 9966413710, e-mail: uppala.divya@gmail.com



Figs 2A and B: Proliferating neoplastic glandular patterns, bland nuclei with minimal cytoplasm and multiple areas of lobular arrangement noted



Figs 4A and B: Neoplastic glandular epithelial cells arranged in a lobular pattern showing the presence of typical bland 'washed out' nuclei. The same section also revealed the arrangement of neoplastic glandular cells in a typical Indian file arrangement

Moreover, the same section also revealed the arrangement of neoplastic glandular cells in typical trabecular fashion (Figs 4A and B).

DISCUSSION

Polymorphous low grade adenocarcinoma involving salivary glands is a distinct but a rare entity which was earlier termed as 'Terminal Duct Carcinoma' by Batsaki et al in 1971 which was later described by Evans and Batsaki in 1984.³ Origin of this neoplasm, according to John J Batsakis in 1980,⁶ Dardick et al 1990⁷ and Eric R Carlson 1986⁸ is believed to be formed from intercalated ducts and reserved cells. According to Vincet et al² and Calmenso et al⁹ the lesion is generally slow enlarging and metastasis is rare involving usually intraoral minor salivary glands.

In the present two cases, the age of occurrence is between 60 and 70 years, slowly enlarging without any metastasis involving the minor salivary glands, palate and buccal mucosa.

Light microscopic evaluation of two cases of polymorphous low grade adenocarcinoma interestingly depicted various architectural pattern of arrangement of epithelial



Fig. 3: Swelling involving the right side of the face (extraoral view)

on the right side. Section stained with H & E revealed the presence of neoplastic glandular epithelial cells arranged in a lobular pattern showing the presence of typical bland 'washed out' nuclei. Some areas also revealed the presence of lobular arrangements with prominent hyaline or mucinous stroma simulating mucoepidermoid carcinoma.

cells which were cuboidal, low columnar and arranged in the form of solid lobules which were the more prominent feature followed by duct like pattern in strands or fascicular type, mucinous or cribriform types.

All the aforesaid findings are in accordance to Evans and J Batsakis,¹⁰ Ragezi and Richard et al,¹¹ James T Castle et al¹² and Steven D Vincet et al.¹³ Therefore, it can be assumed that the above mentioned histopathological features are important diagnostic parameters for polymorphous low grade adenocarcinoma.

CONCLUSION

The indolent nature of this neoplasm mandates conservative surgical excision. The prognosis is usually good although long-term follow-up should be a part of the treatment and management.

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