

## Management of recurrent polymorphous low grade adenocarcinoma: a case report

*Manejo de adenocarcinoma polimorfo recorrente de baixo grau: relato de caso*

### ABSTRACT

The Polymorphous Low Grade Adenocarcinoma (PLGA) is a common minor salivary gland carcinoma. It mostly affects the buccal mucosa and retromolar region, but the palate is an unusual site. As the carcinoma is low grade the recurrence is also uncommon. Our patient had a PLGA initially in the palate which was treated initially through surgical management but had a recurrence after four years. Hence we planned a partial maxillectomy along with level I nodes. Post-operatively follow-up is successful till date. So the management of a recurrent PLGA through careful surgery and post-operative follow –up with prosthetic rehabilitation is discussed in this article.

**Key-words:** Recurrent PLGA; Partial Maxillectomy

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## INTRODUCTION

The term PLGA was suggested by Evans and Batsakis in 1984.<sup>(1)</sup> PLGA is recently described, predominantly as a minor salivary gland carcinoma characterized by cytologic uniformity, histologic blandness, cellular organizational diversity and an infiltrative growth pattern.<sup>(2)</sup> As the term 'Low Grade' indicates its less aggressive biological behavior. It shows female predilection. It usually involves the palate. The other common sites involved are base of the tongue, upper lip, buccal mucosa, retromolar pad region and rarely the major salivary glands.<sup>(3)</sup> We present a case of recurrent PLGA of the palate with regional lymph node involvement 4 years after the first surgery.

## CASE REPORT

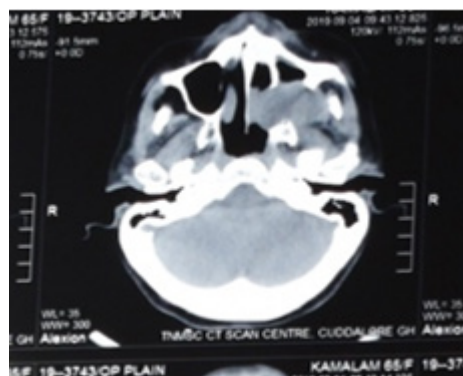
A 67 year old female reported with a chief complaint of pain over the left upper back tooth region for the past 15 days. The pain was continuous and of severe throbbing type which aggravated on eating. She gave a history of previous surgery that was done in her left maxilla before 4 years in our institution. She had proper medical records. She is a case of PLGA that was treated by partial maxillectomy under general anesthesia and immediate reconstruction was done using obturator. She had proper follow up till 2 years post operatively. After that as she was asymptomatic, she didn't show up. On general examination, left submandibular lymph node was palpable measuring about 4 x 3 centimeter that was firm and mobile. She had no other medical conditions.

On examination, a swelling was noticed which was protruding from the anterior margin of the surgical defect (Fig 1). The surface was smooth and no secondary changes were noted. Posterior extent of the swelling couldn't be assessed clinically. On palpation, the swelling was severely tender. Computed tomography (CT) in relation to head and neck was done. The swelling was measuring about 3.9 x 2.7 cm involving the posterior and lateral walls of left maxillary sinus and protruding into the left nasal cavity (Fig 2). Bony changes were noted in the left pterygoid hamulus. A swelling measuring about 2.5 x 2.1 centimeter was noted near the submandibular triangle compressing the submandibular gland suspicious of a lymph node. Fine needle aspiration cytology was performed in the lesion as well as the lymph node. No aspirates could be yield. Incisional biopsy of the lesion was performed. Histologically it predominantly showed necrotic tissue with entrapped degenerated atypical cells in nests and

sheets that gave an impression of a necrotic lesion of the palate. Provisionally the lesion was diagnosed as a recurrent PLGA. impression of a necrotic lesion of the palate. Provisionally the lesion was diagnosed as a recurrent PLGA.



**Figure 1** - Swelling seen through the existing defect



**Figure 2** - CT axial section shows obliteration of the maxillary sinus and nasal cavity

Patient was planned for Partial maxillectomy with removal of level I lymph node on the left side under general anesthesia. Under all aseptic conditions, the patient was prepared. Keen's incision was placed and the lesion was approach through le fort I access osteotomies (Fig 3). Partial maxillectomy was done with a safe margin of 1 centimeter. The entire lesion was removed in toto. Level I Lymph nodes (Fig 4) were assessed and removed by Risdon's incision. The intra oral surgical defect was packed with ribbon gauge soaked in paraffin and Ciprofloxacin ointment. Surgical obturator was placed.



**Figure 3** - Le fort I access osteotomy placed



**Figure 4** - Level I lymph node

The pack was changed every 2 days once in the 1<sup>st</sup> postoperative week. Patient was kept in ward under constant monitoring for the first postoperative week (Fig 5). The wound healing was satisfactory and there were no signs of recurrence till 6 months. Patient was given a definitive obturator and is on regular follow up.



**Figure 5** - Surgical defect seen post -operatively

Histologically, the excisional biopsy of the lesion showed tumor cells with hyalinized stroma. The lymph node showed metastatic tumor cells composed of large vesicular nucleus, prominent nucleolus and having glandular, cribriform and acinar pattern of arrangement. Focal dystrophic calcification was also seen. Based on the histopathology report, the final diagnosis is recurrent PLGA of the palate with regional lymph node metastasis.

## DISCUSSION

PLGA accounts for 19% - 26% of all the minor salivary gland malignancies.<sup>(1)</sup> It mostly involves the minor salivary glands at the junction of soft palate and hard palate. It has female predilection between 60 and 80 years of age.<sup>(2)</sup> The most appropriate choice to manage primary lesion is wide excision with thorough evaluation of the surgical margins. The frequency of local recurrence is 9% - 33%. As the lesion has considerable local recurrence rate, long term follow up is

recommended. In the literature 20 years after the primary surgery, local recurrence was reported.<sup>(3)</sup> This warrants the need for long term follow up. However most of the local recurrence was reported within 5 years of the primary surgery. Studies imply that the majority of recurrences are controlled by surgical re-excision only. Final reconstruction with a microvascular free graft and a fixed or detachable prosthetic reconstruction can be contemplated.<sup>(4)</sup> Therapeutic neck dissections are indicated in cases where positive lymph nodes were assessed clinically or radiographically.<sup>(2)</sup> However prophylactic neck dissections were not recommended.<sup>(5)</sup> The frequency of regional lymph node metastasis ranges from 6% - 35%. Distant metastasis is rare and accounts for less than 1%. Use of radiation therapy is controversial. Some authors suggest radiation therapy for case with lymph node involvement with the primary tumor<sup>(1,5)</sup> Some authors suggest that radiation therapy is indicated in cases with unclear margins, perineural invasion and perivascular invasions. There are no data showing any benefits from adjunctive chemotherapy.<sup>(6)</sup> High grade transformation of PLGA has been reported in cases with multiple recurrence. The possible role of the radiation therapy in initiation of dedifferentiation is still debatable.<sup>(7,8)</sup>

## CONCLUDING REMARKS

This case report highlights the unique clinical presentation of the recurrent lesion (presented as a very painful swelling and had negative margins during the first surgery) and the difficulty in arriving at the provisional diagnosis due to its bland histopathologic features.

It also insist the significance of achieving clear margins for any carcinomas.

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