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RECURRENT POLYMORPHOUS LOW GRADE ADENOCARCINOMA: A RARE CASE PRESENTATION

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ABSTRACT
Polymorphous low grade adenocarcinoma (PLGA) as a malignant salivary gland tumor is difficult to diagnose both clinically & histopathologically due to its dubious presentation and morphologic diversity that includes several microscopic patterns. Biologiccal behavior of PLGA is uncertain, as some cases have developed recurrence & metastasis. A 64-year-old female presented with recurrent polymorphous low-grade adenocarcinoma in left retromolar area, twice during 3years. This case illustrates that despite the indolent clinical behavior of PLGA, long term follow up after treatment is necessary.

INTRODUCTION

Polymorphus low grade adenocarcinoma (PLGA) is a well-recognized malignant salivary gland tumor (MSGT) which was introduced by Evans and Bastakis in 1984 [1]. It occurs predominantly in minor salivary gland and constitutes 5%-11% of MSGTs. The most common site of PLGA is soft or hard palate followed by upper lip and buccal mucosa [2]. Metastatic potential has been reported 5%-15% which is more likely in tumors that recur [3]. We report a case of recurrent PLGA in left retromolar area with previous history of surgery during last 3 years.

CASE REPORT

A 64-year-old female reported in 2014 with a complaint of small mass in left retromolar area which was believed to be second recurrence of left retromolar tumor since initial onset in 2011. She reported first recurrence within 18 months after first surgical procedure. She only had previous pathology reports which representing diagnosis of PLGA with free surgical margins. Intraoral examination showed a 2x1.5cm nontender swelling of left retromolar area with soft consistency and fixation to underlying tissue. The overlying mucosa was erythematous (fig1).

Extraoral evaluation was within normal limits. The computed tomography (CT) examination showed no evidence of bone penetration (fig2). Reevaluation of previous pathologic slides confirmed the diagnosis. So a clinical diagnosis of recurrent PLGA was established. To evaluate the cervical lymph node, CT scan of head and neck was requested but no evidence of pathologic lymphadenopathy was seen.

The patient underwent surgery and histopathological result revealed malignant salivary gland tumor composed of monotonous round to ovoid cells with eosinophilic cytoplasm and also clear cells were arranged in a variety of morphologic pattern including small tubules, solid islands, cribriform, papillary and single file with infiltrative growth to surrounding tissue. Vascular invasion

was also present (fig3). Based on histopathologic findings, history and clinical examination the diagnosis of recurrent PLGA was made. After surgery, the patient is under observe and actually there is no problem during last 6 months.

DISCUSSION AND CONCLUSION

PLGA is a relatively rare malignant salivary gland neoplasm which constitutes 0.6%-7.1% of all salivary gland neoplasm in different geographic distinct. Reviewing of 366 cases of salivary gland tumors in southern iran indicating rarity of the lesion. According to these data. PLGA represents 0.81% of all salivary gland tumors and 2.5% of malignant salivary gland neoplasms [3]. In another retrospective study of 130 cases of salivary gland tumor in Iran by Ansari et al. no case of PLGA was seen [4].

PLGA is the second most common intraoral malignant salivary gland neoplasm after mucoepidermoid carcinoma [2]. However, approximately 32 PLGAs in major salivary glands specially parotid and intraosseous. PLGA have been reported, Either arising denovo or malignant component of carcinoma ex pleomorphic adenoma [5]. Two thirds of tumors have occurred in females with peak prevalence in the sixth to eighth decades of life similar to present case [6-7] but in contrast to most reported cases, our case occurred in retromolar area. The lesions usually manifest as a slow growing and asymptomatic mass which results in diagnostic delay [8].

Fig 1. A small exophytic mass with erythematous surface

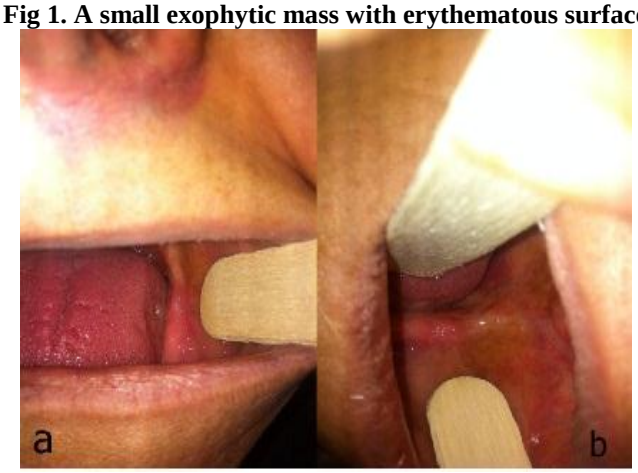


Fig 2. CT image shows no bone penetration

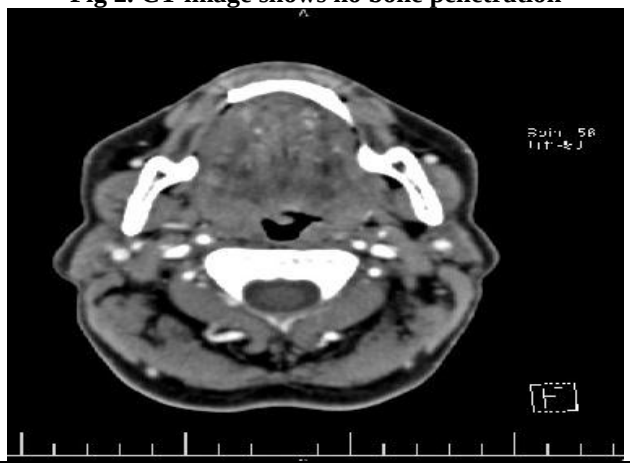
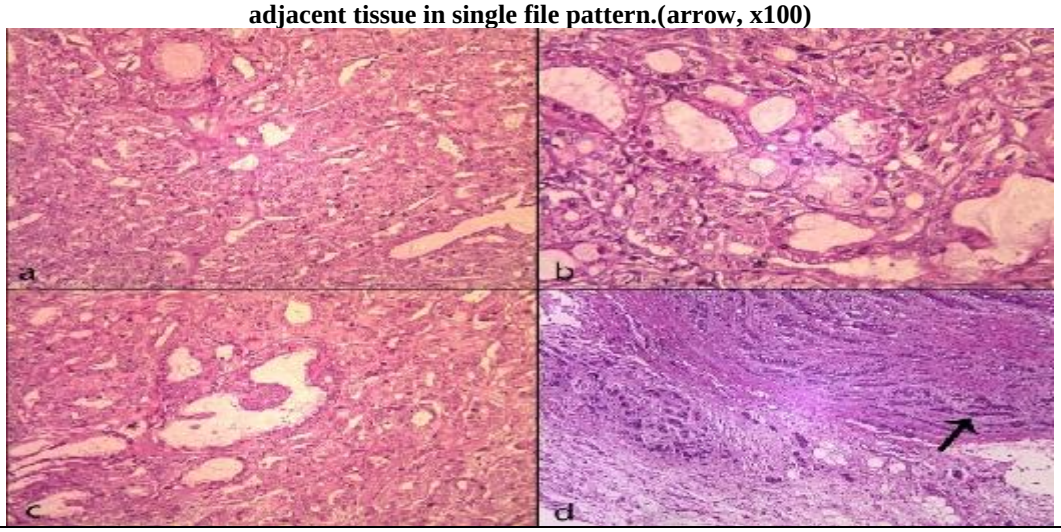


Fig 3. Representing the morphological diversity and cytologic uniformity of PLGA.a) solid tumoral sheet with ductal structures (x200).b)Cribriform pattern of PLGA(x400). c) Papillary projection in cystic space(x400). d) Infiltration to adjacent tissue in single file pattern.(arrow, x100)



Histopathologically, PLGAs are characterized by cytologic uniformity, diversity of cellular organization, histologic blandness and infiltrative growth pattern [9]. Tumor cells are round to polygonal in shape with pale to eosinophilic cytoplasm, arranged in different patterns [10]. Tumor often appears well circumscribed [11]. However, the peripheral cells are usually infiltrative to adjacent tissue in a single-file fashion [12]. Perineural invasion is common [8]. Recurrence, metastasis to regional lymph nodes and distant sites are more likely in the papillary variant similar to our case [11].

Adenoid cystic carcinoma (Adcc) and cellular pleomorphic adenoma should be considered in histologic differential diagnosis [12]. The diversity of growth patterns particularly fasicular areas, micropapillary structures and also lack of basaloid tumor cells help to distinguish PLGA and separation from Adcc. Pleomorphic adenoma lacks an infiltrative border and have focal mesenchyme-like component consisting of myxoid, hyalinized or chondroid background or combination of backgrounds. Foci of plasmacytoidmyoepithelial cells are helpful [6]. Staining with GFAP may be useful with pleomorphic adenoma differential diagnosis. GFAP expressed in 93% to 100% of PAs. In contrast, it is not expressed in PLGA [7]. Recurrence has been reported in 9%-17% of PLGAs, but most of the occurrence has been found after 5 years [10], despite current case which showed twice recurrence during 3 years. Furthermore, Castle *et al*., showed an average time to recurrence of 7.2 years with an average follow up of 115 months [3]. Owing to the possibility of local and regional recurrence, an adequate follow up period of at least 5 years is necessary after initial surgical treatment [7]. Treatment consists of wide local excision with possible adjuvant radiation therapy for inadequate margins or recurrent tumors [10]. However, the 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 only with no recurrence noted [7]. In conclusion, PLGAs are slow growing asymptomatic masses, seen predominantly in palate but recurrence and lymph node metastasis can cause severe problem depending on the tumor site. In these cases adjuvant radio therapy is recommended.

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