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Case Report

Carcinoma ex pleomorphic adenoma in the buccal minor salivary gland: a rare case report

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Abstract: Carcinoma ex pleomorphic adenoma (CXPA) comprise approximately 3.6% of all salivary gland tumors, 12 % of all salivary gland malignancies, and 6.2% of all pleomorphic adenoma variations. The most frequently affected sites are parotid and submandibular gland respectively. In minor salivary gland sites, CXPA occurs most commonly in the palate and it is absolutely rare in the buccal mucosa of oral cavity. CXPA in the cheek has been reported in only a few cases in general population. We present a case of CXPA on the left buccal mucosa in a 48- year-old female. Histopathological findings revealed a pre-existing pleomorphic adenoma which certain areas showed islands of dysplastic epithelial cells. The tumor was non-invasive type.
Keywords: minor salivary gland , pleomorphic adenoma , Carcinoma ex pleomorphic adenoma.

INTRODUCTION

We report a case of carcinoma ex pleomorphic adenoma (CXPA) on the left buccal mucosa in a 48-year-old female. Histopathological findings revealed a pre-existing pleomorphic adenoma which certain areas showed islands of dysplastic epithelial cells. The tumor was non-invasive type. CXPA in the cheek has been reported in only a few cases in general population. Three are three different types of malignant changes in the pleomorphic adenoma (PA), including: carcinoma ex pleomorphic adenoma (CXPA), metastasizing pleomorphic adenoma and carcinosarcoma [1]. Carcinosarcomas are composed of malignant mesenchymal and epithelial components and are also called 'true malignant mixed tumors' and metastasizing PAs are characterized by the existence of one or more metastatic foci but histologically benign PAs, however these are excessively rare [2]. The present case report describe a CXPA that has been originated of a pleomorphic adenoma which an epithelial malignancy is derived. Approximately 3% to 5% of all salivary gland neoplasms and 5% to 15% of all salivary gland malignancies are CXPA[3]. In minor salivary gland areas, CXPA occurs most commonly in the palate while in the buccal mucosa of the oral cavity it is absolutely rare [4]. Due to the low incidence of this condition, no standard modality of treatment has been defined so far. Similar to most other salivary gland malignancies, the main treatment for CXPA is surgery, and post operative radiation therapy plays an important role in the current treatment strategy [5]. We present a case of carcinoma ex pleomorphic adenoma that occurred in the cheek minor salivary gland.

CASE REPORT

A 48- year-old female referred for evaluation of a 20 year history of a painless mass, located in her left cheek mucosa of the oral cavity. There were no accompanied symptoms including pain and tenderness except that it had become increasingly noticeable (Fig-1). An intraoral examination showed a non tender, painless and well circumscried firm movable mass. Covering oral mucosa on the mass was intact and it was not fixed to the surrounding tissues. Expression of

saliva from the left parotid duct was Normal. There were no trismus and palpable regional lymph node. Moreover the function of cranial nerve VII was normal .The patient was healthy and normal regarding systemic clinical examination. Our first clinical diagnosis was lipoma or fibroma or intra mascular hemagioma. MRI of the face with and without contrast medium revealed a well-defined and round mass (32.27.20 mm) in deep part of left buccal area just anterior to masseter muscle with moderate and inhomogenous contrast enhancement (Fig-2). The tumor was resected intraorally under general anesthesia. We approached the lesion via vertical incision in the anterior part of oral cavity because of its large size. After blunt dissection over the intact capsule of the tumor, it was removed totally (Fig3-4). This excisional biopsy revealed a yellow mass that appeared to be well encapsulated. Consistency of specimen was elastic and in cutting was solid. (Fig-5). Histopathologically, the tumor was a combination of salivary gland epithelial and myoepithelial cells seen in a mesenchymal background. The epithelium formed ducts, cystic structures, islands and sheets of cells. In some areas, keratinizing squamous cells were seen. Myoepithelial cells were in rounded, angular, spindled and in some areas plasma cytoid shape. These cells were diffused generally but in focally pattern. Tumoral cells were in a myxoid and fibrotic stroma. In some parts of the tumor, epithelial cells had large nuclei with coarse chromatin and prominent nucleoli. Cytoplasm pleomorphism and mitotic activity of these cells also were seen. (Fig 6-7)

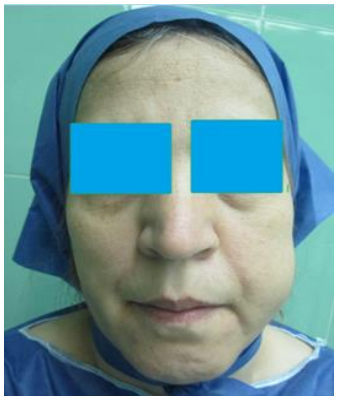

Fig-1: Extraoral view

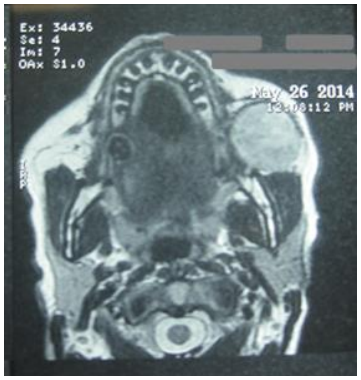

Fig-2A: Axial T1-Weighted MRI

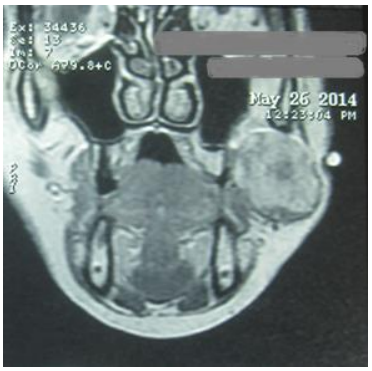

Fig-2B: Coronal T2-Weighted MRI; showing a well-defined and circular mass in deep part of left buccal area just anterior to masseter. This mass showing moderate and in homogenous contrast enhancement

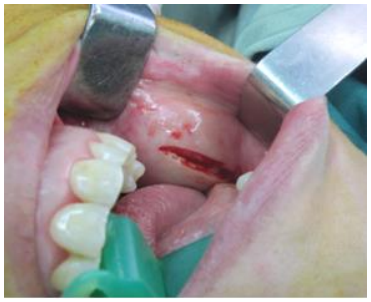

Fig-3: Vertical incision in the anterior part of oral cavity

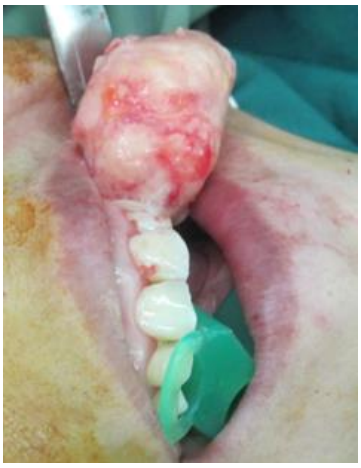

Fig-4: Intraoperative photograph showing an encapsulated mass

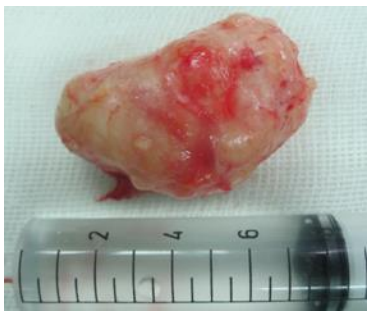

Fig-5: Excised tumor; ovoid and solid

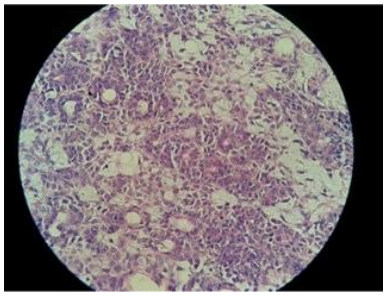

Fig-6: Higher magnification of the PA area in acomposed of glands with myoepithelial cells in a chondromyxoid stroma (H&E, ORIGINAL MAGNIFICATION 40)

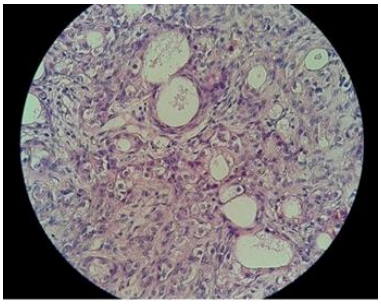

Fig-7: Higher magnification of the carcinoma in a composed of Glandular formation, marked cellular and nuclear pleomorphism and hyperchromatism (H &E, original magnification 40)

DISCUSSION

Among the all minor salivary gland tumors, 40-50% are malignant which are approximately 7% of them are carcinoma ex pleomorphic adenoma [6]. CXPA comprise approximately 3.6% of all salivary gland tumors, 12 % of all salivary gland malignancies, and 6.2% of all pleomorphic adenoma variations .The most frequently affected sites are parotid and submandibular gland respectively. CXPA have been reported more in women with an average 60 years old [4]. The most common clinical presentation of CXPA is a mass with firm consistency in the parotid gland. Pathologic assessment is the gold standard for making a definitive diagnosis [3]. In the minor salivary gland, these lesions most often create in the palate. CXPA from the buccal mucosa is very rare, and only a few cases have been reported till now [4, 7-11]. CXPA has been subclassified as follows:(I)non-invasive ,(II)minimally invasive(<,=1.5 mm penetration of the malignant component into the surrounding tissue),and (III)invasive (>1.5 mm of invasion from the tumor capsule to the adjacent tissue). Groups (I) or (II) usually have an excellent prognosis but group (III) exhibit significant recurrency and metastasis[4,12]. In the present case ,our final diagnosis was non-invasive carcinoma ex pleomorphic adenoma that also known in the literatures as intracapsular CXPA and carcinoma in situ, intratubular or Intraductal CXPA [13] .Unfortunately, malignant changes in pleomorphic adenoma occurs most frequently in the lesions with long-standing time or lesions with multiple local recurrences. The risk of malignancy increases with the duration of the mass. Only 1.6% of malignant transformation occurs in tumors less than 5 years, while it is approximatly 9.5% for tumors over 15 years [14]. Therefore adequate removal of this carcinoma ex pleomorphic adenoma is critical. Although metastasis to lung is rare, patient treated for carcinoma ex pleomorphic adenoma should be investigated for distant metastasis, such as lung and bone [5], thus examination and long term follow–up are necessary for local recurrency and systemic metastasis. Treatment of CXPA often involves a radical surgical procedure which may be followed by radiotherapy [3]. Currently, Surgery is the primary choice of treatment for CXPA [5]. In some studies, postoperative radiotherapy was recommended for CXPA with advanced stage, lymph node involvement, high histological grade as well as perineural invasion; however surgery alone may be a choice option for small and non-invasive carcinomas similar to our case. It should be noted that further studies are necessary to ascertain which patients can really benefit from postoperative radiotherapy [15].

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