

CASE REPORT

Pleomorphic Adenoma of Minor Salivary Glands: a Rare Case Report

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ABSTRACT

Pleomorphic adenoma is a non-cancerous neoplasm that develops in the salivary glands. Originating from minor salivary glands, it is extremely uncommon and primarily affects females. The peak incidence is observed between 40 and 60 years of age. It usually presents as a slowly growing, painless, solid tumor that does not cause ulcers on the overlying mucosa. Here, a 47-year-old woman experienced repeated swelling on the buccal mucosa following surgical extraction of a pleomorphic adenoma three years ago. The swelling was solid with clearly defined boundaries. An excisional biopsy was conducted under general anesthesia, resulting in total mass removal. The histological evaluation revealed the existence of a recurring pleomorphic adenoma. This instance emphasizes the significance of addressing this entity as a potential etiology for persistent painless and intraoral swellings.

Keywords: salivary gland neoplasms, minor salivary glands, pleomorphic adenoma, recurrent cheek mass, diagnosis, therapy.

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Article received on the 4th of March 2024 and accepted for publication on the 17th of May 2024

INTRODUCTION

Salivary gland neoplasms are a diverse collection of tumors that display various clinical, histological and immunohistochemical characteristics (1). These tumors are uncommon, comprising only 3% to 5% of all neoplastic entities that occur in the head and neck area (2). Out of these, around 64.9% to 67.5% are benign tumors (3, 4). Neoplasms originating from minor salivary glands present in the mucosa of the cheek, lip and tongue are exceptionally uncommon (3%-5%). Most minor salivary gland tumors are cancerous.

Pleomorphic adenomas (PAs) are the most prevalent type of benign tumors of the salivary glands, comprising 33.2% to 68.4% of all reported cases (4-6). The yearly occurrence is roughly 2-3.5 instances per 100,000 individuals. It manifests in individuals across all age groups (7). However, it is predominantly prevalent among females, accounting for 60% of cases, particularly in the age range of 40 to 50 years (4, 5, 8, 9). The occurrence of these tumors is observed in both major salivary glands (65%) and minor salivary glands (35%). For the majority of cases (70-80%), this tumour primarily occurs in the superficial lobe of the parotid gland, while in about 10% of cases it is found in the submandibular salivary gland, and only 1% in the sublingual gland (10). Less commonly affected areas include the cheek mucosa, palate, lip and tongue, which are accounting for 5-10% of cases (2, 4, 5, 8). From a clinical perspective, PA manifests as a gradually developing, painless abnormality, with a solid texture and varying sizes (11, 12), but with clear boundaries. The likelihood of PA developing malignancy is approximately 6%. The precise cause of pleomorphic adenoma is still unknown, although long-term exposure to radiation and the presence of simian virus (SV40) may be significant factors. Previous studies have documented clonal chromosomal abnormalities characterized by aberrations affecting the 8q12 and 12q15 regions (13).

The intraoral PA is typically situated in the submucosa and has a stiff or rubbery texture. The mucosal lining remains untouched, but ulcerations are present in certain instances (11, 12). Within these specific areas inside the mouth, such as the cheek mucosa, lip and tongue, which are prone to injury, PA might exhibit similar clinical

characteristics to other reactive inflammatory diseases of the salivary glands as well as noncancerous growth processes. In light of this, we provide a case of recurrent PA in the cheek mucosa after surgical removal of pleomorphic adenoma three years ago. □

CASE REPORT

A 47-year-old female presented to the outpatient section of the Maxillofacial department of our hospital with a painless swelling in her left cheek that has been present for more than six months (Figure 1). Thorough history and evaluation revealed that the swelling initially appeared as a little mass and has progressively grown in the same area, where she had been operated for a pleomorphic adenoma (PA) of minor salivary glands three years ago. No lymph node involvement was observed during the extraoral physical examination. An intraoral examination revealed a sizable, non-painful, movable mass in the mucosa of the left cheek, measuring approximately 5 cm in diameter. The lesion exhibited clear boundaries, displayed a normal color and texture of the overlying mucosa. Furthermore, there was inability to open the mouth and generally there was malfunction to the mobility of oromandibular system. The motor and sensory nerve functioning were undamaged. During bimanual palpation, the mass was detectable in the space between the skin of the cheek and the mucosa. There were not accompanying symptoms. There was no previous medical history of hypertension or diabetes, but she was a heavy smoker. There were no reports of head and neck tumors in her family history.



FIGURE 1. Painless swelling in the left cheek

Based on clinical data, the diagnostic possibilities included fibroma, lipoma or inflammatory fibrous hyperplasia due to the region's susceptibility to trauma. However, the possibility of an inflammatory process can be eliminated, as there

was no evidence of infection in this area. The differential diagnosis includes mucoepidermoid carcinoma, adenocystic carcinoma, tumor of the accessory parotid salivary gland, epidermoid cyst and dermoid cyst. Initially, we had to consider the possibility of pleomorphic adenoma recurrence based on the patient's medical background. The patient underwent ultrasound and magnetic resonance imaging (MRI) of the head and neck. In the anatomical position of the masseter muscle on the left side, a clearly defined inhomogeneous (with solid and cystic elements), hypoechoic mass with lobular borders and central vascularization on color Doppler, measuring 4 x 4 x 3.6 cm, was seen (Figure 2). No abnormally enlarged lymph nodes and infiltration of adjacent bony structures were identified. The magnetic resonance imaging revealed a clearly delineated, moderately-enhancing region located in the left cheek, in contact with the masseter muscle.

Fine needle aspiration (FNA) with local anesthetic indicated the presence of pleomorphic adenoma. Following the essential preoperative assessments, a horizontal incision was performed on the mucous membrane of the cheek, parallel to the occlusal plane, while the patient was under general anesthesia. The parotid duct was observed and circumvented. The lesion was located within the space between the mucosal and the buccinator muscle while diligent hemostasis was performed. It was readily separated from the adjacent tissues during dissection. The mucosa was stitched and the sample was sent for histological examination. The tumour size was 4 x 4 cm (Figures 3 and 4).

The patient's healing and recovery proceeded smoothly, with no indications of recurrence during the one-year follow-up period (Figures 5 and 6). The authors acquired the patient's explicit written approval to publish this material, adhering to the ethical requirements.

From a histopathological perspective, the tumor mass was clearly surrounded by a well-defined capsule. The epithelial cells exhibited a configuration characterized by a duct-like pattern, cystic formations and anastomosing cords. The specimen exhibited a fragile collagenous framework with regions displaying a gelatinous and cartilaginous composition. Every characteristic indicated the presence of pleomorphic adenoma. □



FIGURE 2. A clearly defined hypoechoic, inhomogeneous mass with lobular borders and central vascularization on color Doppler

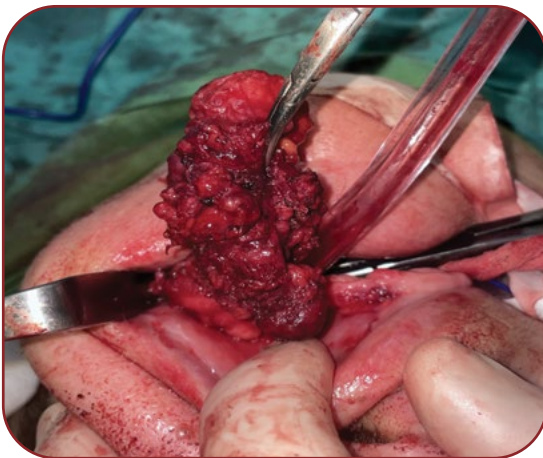


FIGURE 3. Intraoral removal of the tumor en block

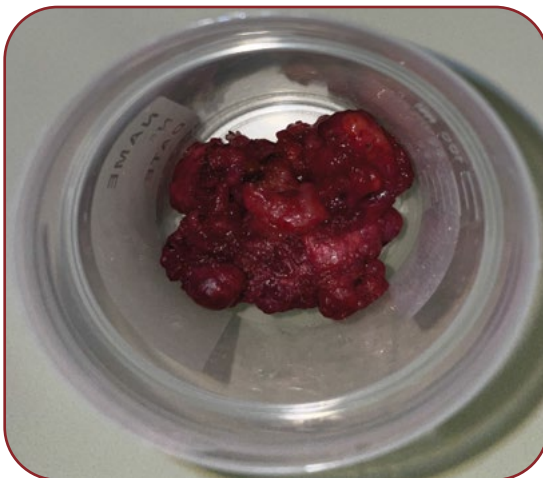


FIGURE 4. Surgical specimen



FIGURES 5 AND 6. Smooth wound healing and no recurrence one year postoperatively

DISCUSSION

The buccal area is highly intricate and encompasses a wide array of components. The differential diagnosis of a buccal space lesion encompasses the possibility of a tumor arising from several tissues, such as glandular, vascular, lymphatic, connective, muscular, ductal and neural structures (14).

Pleomorphic adenoma primarily affects the large salivary glands (4, 9, 12, 15) and is seldom in the minor salivary glands (2, 9, 12, 15, 16). Therefore, the current instance of PA in the minor salivary glands of the cheek mucosa validates the existing literature that indicates a relatively low occurrence in this location (2, 16) compared to other sites like the palate (64%) and less frequently afflicted areas such as the upper lip (13%) (8). The epidemiologic data of PA aligns with the literature, indicating that the gender and age of the current case are consistent with the majority of cases. It is commonly observed that PA in the minor salivary glands predominantly affects women in their forties and fifties, with an average age range of 45-50 years (4, 5, 8).

Fine-needle aspiration cytology (FNAC) is considered the most accurate method for dia-

gnosing PA. Nevertheless, PAs may erroneously be identified as low-grade cancers. This discrepancy is uncommon in the literature. Hence, if there is any doubt, it is advisable to have a guided FNAC procedure, ideally with confirmation from two distinct pathologists. Some locations, such as tiny tumors near the jaw, are challenging to evaluate with FNAC. In such cases, an incisional biopsy may be necessary due to the diverse nature of salivary gland tumors (17).

Pleomorphic adenomas originating from the minor salivary glands exhibit a low likelihood of recurring, with recurrence rates ranging from 2% to 44%. The recurrence of surgical issues is mostly attributed to pseudopodia, capsular penetration, tumour rupture and inadequate surgical technique. It is imperative to perform dissection of salivary gland tumors due to their inherent tendency to develop into malignancies. The initial therapeutic approach should be wide local excision with negative margins (7) and preservation of the capsule (11, 12, 16, 18, 19). It is imperative to choose the appropriate surgical procedure in order to prevent recurrence. Early detection of PA is crucial due to the potential for secondary malignant change into carcinoma ex pleomorphic adenoma, despite the low incidence of the tumour (9, 15, 18, 20). □

CONCLUSION

The current findings underscore the need of accurately diagnosing and treating the abnormalities that may impact the oral and maxillofacial areas as well as the need for continual evaluation of cases, especially those identified as PA. Early diagnosis is crucial as it allows appropriate treatment of the lesion and improves the prognosis. Regarding this matter, PA necessitates extended observation to examine potential recurrences and/or progression of the case. □

Conflicts of interest: none declared.

Financial support: none declared.

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