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Case Series

Pleomorphic Adenoma of Minor Salivary Glands: A Report of Three Cases and Literature Review

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Abstract: ***Introduction:*** Pleomorphic adenoma is the most common benign tumor of the minor salivary glands. Despite its benign nature, its potential for recurrence and malignant transformation makes it clinically significant. This study aims to describe the epidemiological, diagnostic, and therapeutic characteristics of pleomorphic adenomas of the minor salivary glands and to compare the findings with those reported in the literature. *Materials and* ***Methods:*** A retrospective descriptive study was conducted over a two-year period (March 2023 to March 2024), involving three patients diagnosed with pleomorphic adenoma of the minor salivary glands. Clinical features, diagnostic imaging, treatment approaches, and follow-up data were collected and analyzed. ***Results:*** The study included three patients (1 male, 2 females) with a mean age of 22.7 years. Tumor locations included the hard palate, lower lip, and inner cheek. The average time to diagnosis ranged from 1 to 6 months. In all cases, the clinical presentation was a painless swelling. Diagnostic imaging included soft tissue ultrasound and CT scan. Surgical excision was performed under local anesthesia using a monobloc technique. Histopathological analysis confirmed the diagnosis of benign pleomorphic adenoma in all cases. One recurrence was observed after 18 months of follow-up. ***Conclusion:*** Pleomorphic adenoma is the most frequent histological subtype of benign minor salivary glands tumors and typically exhibits slow progression. It should be considered in the differential diagnosis of any painless, submucosal lesion in the oral cavity. Complete surgical excision with a 5 mm margin of healthy tissue remains the treatment of choice. Due to the risks of recurrence and malignant transformation, long-term follow-up is essential.

Keywords: Pleomorphic adenoma, minor salivary glands, oral cavity, hard palate, surgery, recurrence.

INTRODUCTION

Tumors of the minor salivary glands are rare and are most often benign. However, the most frequently encountered histological type—pleomorphic adenoma (PA)—remains a major concern for maxillofacial surgeons due to its potential for malignant transformation. Clinical and paraclinical presentations are variable. Management is primarily surgical, requiring wide and complete excision of the tumor. Histopathological examination confirms the diagnosis, and long-term follow-up is essential. The objective of this study was to describe the epidemiological, diagnostic, and therapeutic

features of pleomorphic adenoma of the accessory salivary glands.

MATERIALS AND METHODS

We report a retrospective study of three cases of pleomorphic adenoma of the minor salivary glands managed between March 2023 and March 2025 in the Department of Stomatology and Maxillofacial Surgery at Ibn Tofail Hospital, Mohammed VI University Hospital Center in Marrakech MOROCCO. Medical records were reviewed retrospectively. All patients underwent clinical examination and radiological assessment, were operated on by the same surgical team, and their surgical specimens were analyzed by the same pathology laboratory.

RESULTS

The study included three patients: one male and two females. The mean age was 22.66 years, with ages ranging from 14 to 35 years.

The first case involved a 35-year-old female presenting with a hard palate tumor. She consulted for a painless palatal swelling (Fig. 1), approximately 1 cm in diameter, firm, well-defined, and covered with normal-appearing mucosa. The lesion had been evolving over an average period of 6 months. The remainder of the cervicofacial examination was unremarkable.

Computed tomography (CT) revealed a homogeneous soft tissue density mass in the hard palate, with well-defined, regular margins and moderate contrast enhancement. No underlying bone destruction was observed.

The second case involved a 14-year-old adolescent girl who presented with a swelling of the left cheek (Fig. 2), with an average evolution period of one month. The lesion was located on the inner aspect of the cheek.

Soft tissue ultrasound revealed an oval-shaped mass within the subcutaneous fat of the left buccal region, measuring 4.4 × 11 mm. The lesion had well-defined margins and a homogeneous hypoechoic echotexture, with no significant vascularization on color Doppler imaging. It preserved the surrounding fat, underlying musculature, and maxillary bone (Fig. 3).

Figure 1: Pleomorphic adenoma of the hard palate

Figure 2: Pleomorphic adenoma of the inner cheek

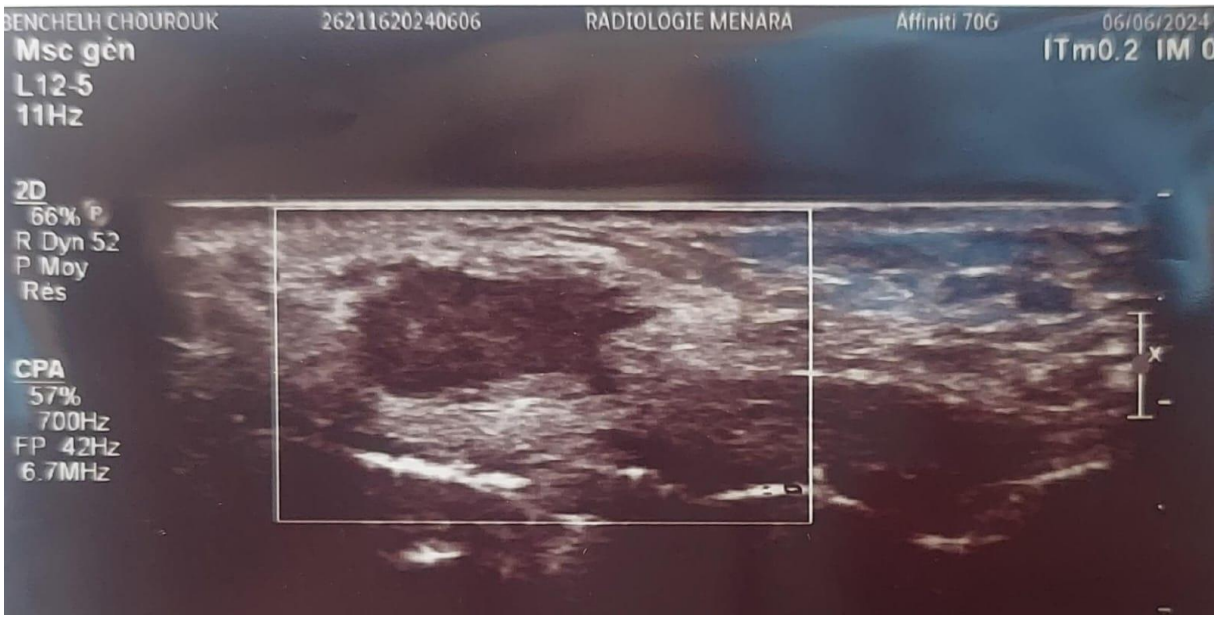

Figure 3: Soft tissue ultrasound showing an oval-shaped lesion measuring 4.4 × 11 mm, with well-defined margins and a homogeneous hypoechoic echotexture, without detectable vascularization

The third case involved a 20-year-old male with a lesion located on the lower lip, presenting as a firm, well-defined nodule on the labial mucosa. The average duration before diagnosis was approximately three months.

Surgical excision of all three lesions was performed under local anesthesia via an intraoral approach, given their small size (not exceeding 1 cm in greatest dimension) (Fig. 4). The patient with the palatal lesion underwent an excisional biopsy, including the overlying palatal mucosa, without a 5 mm safety margin.

Histopathological examination confirmed the diagnosis of pleomorphic adenoma in all cases. The tumors were benign, well-encapsulated, and completely excised.

The mean follow-up period was 1.5 years. One case of recurrence was observed eight months postoperatively in the patient with the palatal localization.

Figure 4: Intraoperative view of a pleomorphic adenoma with complete excision of the tumor and the overlying buccal mucosa, with 5 mm safety margins

DISCUSSION

Tumors of the minor salivary glands (MSG) account for 15–20% of all salivary gland tumors [1,2]. Pleomorphic adenoma (PA) is the most common tumor type, representing around 50% of both major and minor salivary gland tumors [3]. Approximately 80% of PAs arise in the parotid gland, 8% in the submandibular gland, and 7% in the MSG. Among benign MSG tumors, PA is the most frequent

histological type, accounting for 70.6% to 100% of cases, with a predilection for the palate [4].

PA of the minor salivary glands occurs more frequently in women, with sex ratios varying across studies from 1/1.1 [5] to 1/3.2 [6]. In our series, the sex ratio was 0.5. The peak incidence is typically between 30 and 40 years of age [7]; in our cases, the mean patient age was 22 years.

Clinical symptoms depend on tumor size and location [8]. In the oral cavity, PAs generally present as painless swellings beneath normal mucosa [9,10]. Palatal lesions usually affect the posterolateral region, confined between the hard palate and thick fibromucosa, with malignant tumors sometimes causing a "watch-glass" deformation. PAs of the soft palate may present as nodules embedded in the hemivelum, occasionally deviating the uvula or involving the anterior tonsillar pillar. Labial PAs typically appear as firm, well-circumscribed nodules close to the mucosa, with late bulging toward the skin surface.

Clinical findings are often minimal, as these tumors are slow-growing and usually discovered once they become prominent. In our series, all patients presented early, and none of the lesions exceeded 1 cm in greatest dimension. The mean diagnostic delay was three months, ranging from 1 to 6 months.

MRI findings vary depending on the cellular and myxoid composition of the tumor. PA often appears lobulated, well-demarcated, hypointense on T1-weighted images and hyperintense on T2-weighted images, with homogeneous enhancement after contrast injection [11]. However, atypical features such as heterogeneous T2 signal and absence of lobulation may complicate the diagnosis [12,13].

Fine-needle aspiration (FNA), which was not performed in our patients, offers a rapid and reliable diagnostic method when interpreted by an experienced cytopathologist. Reported sensitivity ranges from 73% to 93%, and specificity from 85% to 98% [14]. FNA is cost-effective and may reduce unnecessary surgeries when benignity is confirmed preoperatively, potentially lowering care costs by 25% per patient [15].

Intraoperative frozen section examination, although controversial in Anglo-American settings, remains valuable in distinguishing benign from malignant salivary gland tumors. With a sensitivity of 74%, specificity of 99%, false-negative rate of 3.5%, and false-positive rate of 0.83%, it is a reliable technique especially when preoperative diagnosis is unavailable. However, due to the architectural polymorphism of these tumors, subtyping remains challenging. Frozen sections are also useful in assessing surgical margins [16].

Incisional biopsy is generally discouraged due to risks of recurrence and tumor spread [17]. Clinically, the differential diagnosis includes all benign oral mucosal tumors with a nodular appearance, such as:

Connective tissue tumors: fibroma, lipoma, myxoma

Developmental anomalies: ectopic thyroid nodule, thyroglossal duct cyst, dermoid and epidermoid cysts, lymphoepithelial cyst

Muscle tumors: leiomyoma, rhabdomyoma

Nerve tumors: schwannoma, neuroma, traumatic neuroma, granular cell tumor (Abrikossoff tumor)

Macroscopically, the tumor appears as a nodular, well-circumscribed or encapsulated mass, usually gray-white in color and sometimes translucent. Its consistency varies from firm to gelatinous. The term “pleomorphic” refers to the architectural diversity, in contrast to the cytologic monomorphism of epithelial and myoepithelial cells. A key diagnostic feature is the presence of a myxoid stroma, sometimes with cartilaginous or osseous differentiation [18,19]. Highly cellular areas referred to as “cellular pleomorphic adenomas” may also be seen. A capsule is usually present, except in myxoid-dominant tumors or those located in the oral or nasal cavity [20]. Capsular rupture, incomplete capsules, pseudopodia, and satellite nodules are frequent [21], but these features are not in themselves criteria of malignancy. However, they complicate the assessment of complete resection and may favor local recurrence.

Surgical excision remains the treatment of choice, regardless of tumor location. Enucleation is not considered an adequate approach. Direct incisions with mucosal flap elevation are discouraged due to the risk of leaving tumor islands in the mucosa. Tumor resection often requires sacrificing the overlying mucosa to minimize recurrence, which is influenced by tumor location and surgical completeness [22]. Reported recurrence rates range from 2.4% to 10% [23]. In our series, recurrence occurred in one patient who had undergone complete excision without the recommended 5 mm safety margin.

Malignant transformation—carcinoma ex pleomorphic adenoma (CXPA)—is rare, occurring in fewer than 7% of minor salivary glands PAs, most

commonly in the palate [24]. Risk increases with recurrence frequency and diagnostic delay, from 1.6% before 5 years to 9.4% after 15 years [25]. Clinical signs suggestive of malignant transformation include rapid growth, ulceration, infiltration, and spontaneous bleeding. Histological examination must be thorough, as malignant foci may be minimal [26]. The malignant component often corresponds to adenocarcinoma or undifferentiated carcinoma.

Metastasizing pleomorphic adenoma refers to a histologically benign PA that nonetheless spreads locoregionally or distantly. This phenomenon appears linked to multiple recurrences and/or repeated surgeries that enable vascular dissemination [27]. Secondary locations—typically bone, lungs, and lymph nodes—can appear up to 55 years after the initial diagnosis [18,19].

CXPA is a carcinoma arising from a preexisting PA and may present with any histologic subtype [28]. It can be difficult to diagnose, particularly intraoperatively, as the malignant component may be subtle. Extensive sampling is required to avoid missing transformed areas. Differentiating CXPA from a cellular PA can be challenging, especially when the carcinoma is well-differentiated. While immunohistochemistry is not useful for PA diagnosis, it can be helpful in CXPA to assess the proliferative index of the malignant component.

Management of CXPA is not yet standardized. Wide surgical excision with clear margins is essential for favorable outcomes. In the oral cavity, this often requires mucosal sacrifice and, depending on tumor extent, may involve partial or complete resection of the mandible, hard palate, or soft palate. For sinonasal cancers, partial or total maxillectomy with prosthetic rehabilitation may be necessary [29].

Postoperative radiotherapy is reserved for aggressive tumors those with invasive carcinoma, perineural spread, bone involvement, lymph node metastasis, or incomplete resection [30]. Radiotherapy may also serve as a primary treatment in inoperable cases due to location or extent [13].

CXPA is locally aggressive and recurs in up to 50% of cases, particularly in palatal locations [31]. Distant metastases typically involve the lungs, bones, and brain [31,32]. Recurrence and metastasis are associated with poor prognosis, while non-invasive and minimally invasive forms carry excellent outcomes. Ten-year survival rates range from 18% to 50% [33].

None of our patients showed malignant transformation.

CONCLUSION

Based on this study and literature review, pleomorphic adenoma of the minor salivary glands should be considered in the differential diagnosis of any submucosal mass in the oral cavity, particularly in the palate, under clinically healthy mucosa. Incisional biopsy is not recommended due to the risk of recurrence and tumor spread. Complete surgical excision with a 5 mm margin of healthy tissue remains the treatment of choice. Long-term and rigorous follow-up is essential due to the risks of recurrence and malignant transformation.

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