



Pleomorphic Adenoma of Palatal Minor Salivary Glands with Bone Infiltration: Case Report with Long-term Follow-up

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Abstract

Intraoral pleomorphic adenoma is a rare entity, with the most common site of appearance being the palate. As a benign lesion, it rarely involves underlying structures. The aim is to present a case of a palatal pleomorphic adenoma (cellular) with bone involvement, with reference to clinical and histological features of the tumour, emphasis on diagnostic and therapeutic management and comparison with the findings of similar studies in the literature. Pleomorphic adenomas of the palatal minor salivary glands remain a rather rare clinical condition, and it is unusual to infiltrate the adjacent palatal bone. A thorough clinical imaging and histological/cytological examination should always be performed to define the benign or malignant nature of the lesion, preoperatively.

Keywords Minor salivary gland · Palate · Pleomorphic Adenoma · Bone infiltration · Benign lesion · Surgical excision

Introduction

The palate is the most common site for intraoral pleomorphic adenoma (PA) with a female predilection and a distribution from 20 to 79 years of age (Wu, 2019).

Pleomorphic adenoma is the most common benign tumour of the salivary glands. Major salivary glands, especially the parotid, are the most common site, while minor salivary glands can attribute to 7% of the affected glands.

Clinically, it is usually presented as a firm, painless submucosal mass, slowly increasing in size with age. Occasionally, mucosal ulceration may be present [1]. The delay of diagnosis and management is highly attributed to their indolent, asymptomatic nature.

The aim of this paper is to present a case of palatal cellular pleomorphic adenoma of a minor salivary gland with the relatively rare finding of the underlying bone involvement.

Case Presentation

A 71-year-old female patient was referred to our outpatient clinic. She reported a left-sided palatal lump, first appearing 15 years ago, with gradual growth.

Clinical intraoral examination revealed a sizeable well circumscribed projecting mass of her left side posterior palatal mucosa (Fig. 1). The overlying mucosa was normal with no evidence of any ulceration. The mass was firm and painless to palpation.

CT scan revealed a 30 × 30 × 40 mm osteolytic lesion involving the left palatal and maxillary alveolar bone, invading to the ipsilateral maxillary sinus and nasal cavities with well-defined peripheral boundaries, alike benign lesions (Fig. 2).

The lesion underwent an FNA cytologic examination preoperatively with no signs of malignancy.

The patient was then scheduled for a pericapsular tumour excision with a mild extension of bone cuff resection. The tumour was excised after local palatal plus vestibular mucoperiosteal flaps reflection under general anaesthesia (Fig. 3). The adjacent alveolar bone and molar fragment was also

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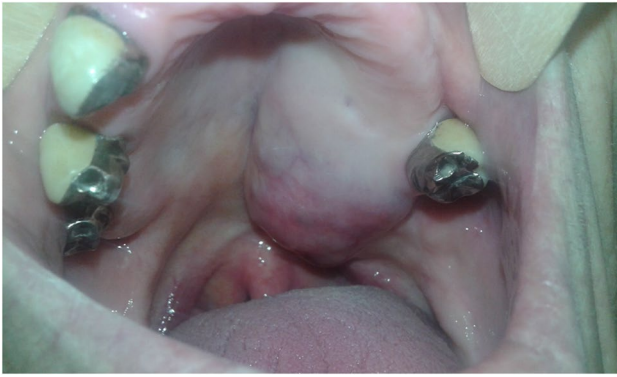


Fig. 1 Clinical image of the lesion. A sizeable well-circumscribed projecting mass of her left side posterior palatal mucosa

resected on surgical specimen (Fig. 4). The intraoperative subsequent oro-antral communication was closed via first aim surgical wound closure. Postsurgical period was uneventful, and the patient followed a regular 5-year clinical and imaging follow-up schedule (Fig. 5). Six years after resection, no recurrence evidence has occurred.

The surgical specimen was sent for pathological examination. The final diagnosis was that of cellular pleomorphic adenoma of palatal minor salivary glands with extension to the underlying bone.

Microscopically, the lesion corresponded to a neoplasm of relatively high cellularity composed of columnar and

cuboidal cells with epithelial and myoepithelial features. The neoplastic cells comprised eosinophilic or clear cytoplasm and oval or roundish nuclei, without any significant pleomorphism. The architectural pattern of the epithelial/myoepithelial component was tubular, glandular, and cribriform, in cords, islets, and groups. The stroma was fibrous, hyalinized with sparse psammoma bodies. Mitoses were rare. The periphery of the lesion was surrounded by hyalinized, fibrous band, with the presence of lobules of minor salivary glands (Fig. 6).

Discussion

Pleomorphic adenoma is considered the most common benign tumour of all salivary glands. Accounting minor salivary glands, more than 80% of arising tumours are malignant [2].

In a case series published by Vicente et al., the hard palate is the most common site of pleomorphic adenoma in minor salivary glands, comprising almost half of all cases (42.5%). Other intraoral sites are less common, such as lip mucosa, soft palate, and buccal mucosa. Extraorally, head and neck pleomorphic adenomas have been described to be in the sinuses or the pharynx and larynx. Even a pulmonary pleomorphic adenoma has been described in the literature [3].

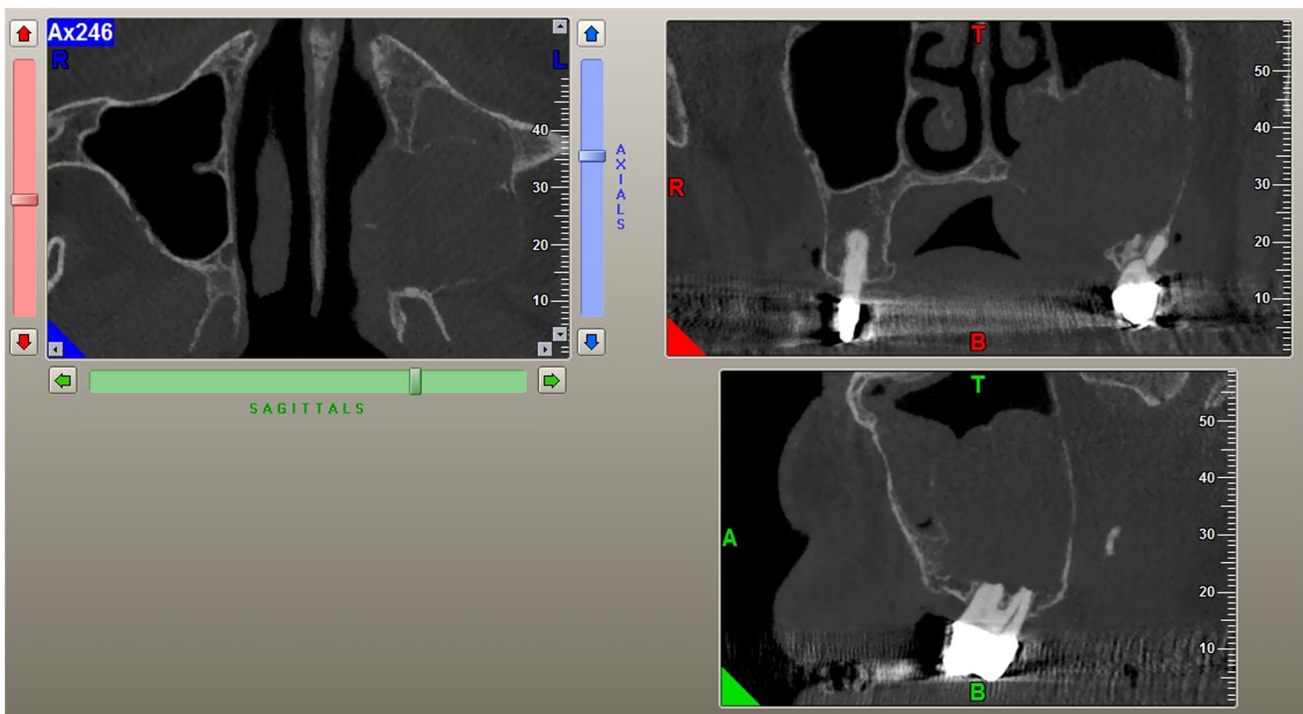


Fig. 2 CT scan; axial, coronal and sagittal view. An osteolytic lesion involving the left palatal and maxillary alveolar bone

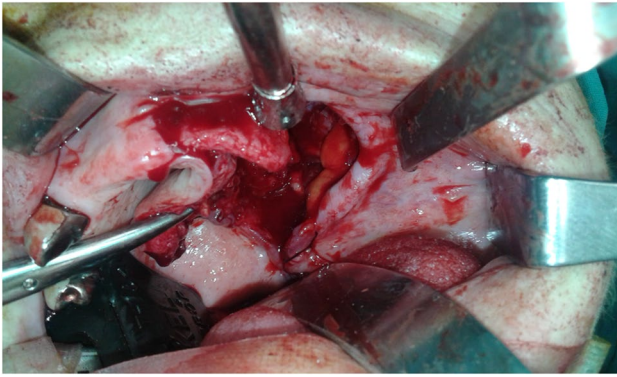


Fig. 3 Intraoperative image of the raised flaps and the lesion



Fig. 4 Surgical specimen, the lesion was excised attached to the alveolar bone



Fig. 5 The 5-year follow-up shows no sign of recurrence

A combination of proper preoperative imaging and cytology/histopathology testing should be evaluated, favouring the definition of the management plan.

FNA cytology is a routine procedure for salivary lesion identification. Research shows a high sensitivity but low specificity in benign tumours, and thus, the evaluation of the result in pleomorphic adenoma is considered controversial.

Needle core biopsy technique provides higher specificity compared to FNA cytology, being sometimes able to insinuate possible metastatic origin of the tumour.

CT and MRI scans are used for the definition of the lesion's size, type of boundaries, and possible expansion to adjacent tissues and structures. In our case, CT (CBCT in particular) was preferred since the clinically suspected bone erosion would be better evaluated, compared to MRI.

In our case, based on the benign clinical features of a chronic increasing in size lesion, accompanied by imaging findings of well-defined peripheral boundaries plus preoperative FNA biopsy negative result for malignancy, we performed a pericapsular tumour excision with a mild extension of bone cuff resection.

In general, bone infiltration is considered a rather rare complication of benign tumours, and especially pleomorphic adenomas. A recent study of case series from India, describes 4 patients with full thickness erosion of the maxillary bone, treated with wide excision [4]. After 1 year of follow-up, no recurrence was recorded. Another recent study of case series records a 66.7% rate of patients (12 out of 18) with palatal pleomorphic adenomas, without evident bone infiltration [1]. No recurrence was also recorded in a 4-year follow-up, excluding a patient with a ruptured tumour intraoperatively, who obviously had a recurrence. A large case series, studying 74 palatal pleomorphic adenomas, does not record any bone involvement [5].

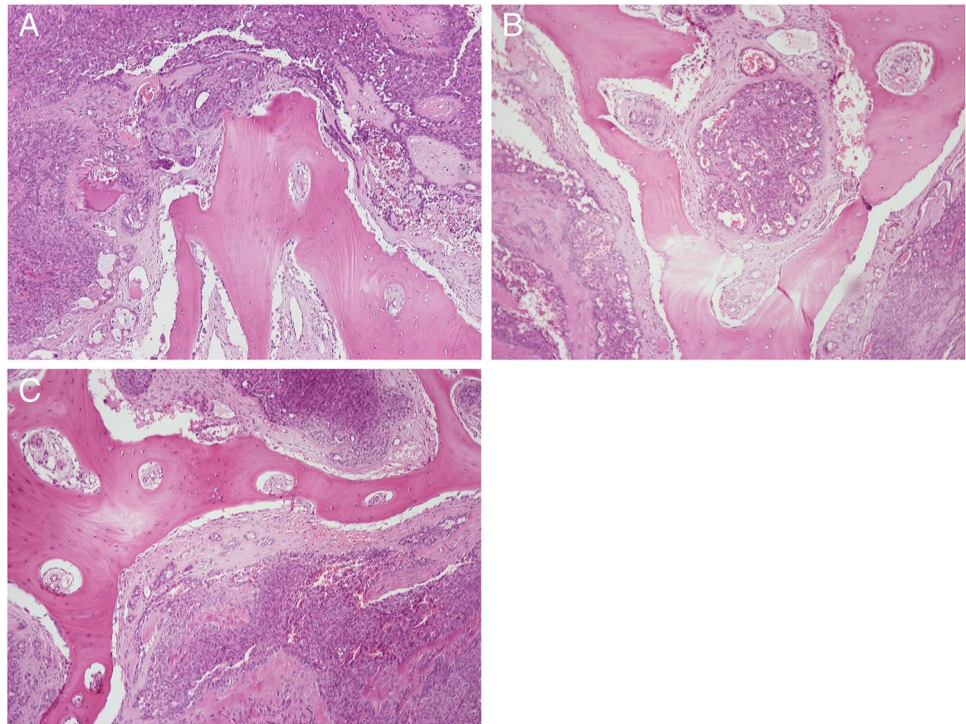
A possible theory that may arise from our case report is that a chronically evolving (long standing) tumour may erode the adjacent bone in such an extent that its total destruction by tumour infiltration can be the result.

A long interval between the appearance of first symptoms and diagnosis, as in our case, contributes to bone erosion and further infiltration due to mechanical pressure by the tumour.

Differential diagnosis, in the first place, comprises entities that are encountered at minor salivary gland sites sharing common features with the given tumour. Tumours that almost exclusively affect minor salivary glands are low-grade polymorphous adenocarcinoma, cribriform adenocarcinoma of minor salivary glands, clear cell carcinoma, and canalicular adenoma. On the other hand, mucoepidermoid carcinoma and adenoid cystic carcinoma still may affect both major and minor salivary glands [6]. In PAs, squamous metaplasia sometimes is the result of FNA or trauma and should be evaluated with caution in the differential diagnosis with SCC or MEC.

On histological grounds, PAs show a broad range of morphological patterns with the main feature being that of a biphasic tumour, composed of luminal ductal and abluminal myoepithelial components of varying proportions [7]. Architectural structures of neoplastic cells, as well as the stroma, may show large morphological variety. Depending on the prevailing histologic elements of

Fig. 6 Pathology specimen. **A** Osseous trabeculae with infiltration by the neoplastic cells of cellular pleomorphic adenoma (arrows). **B** A high cellularity tumour composed of columnar and cuboidal cells with epithelial and myoepithelial features, surrounded by bone with inflammatory lesions. **C** The neoplastic cells comprised eosinophilic or clear cytoplasm and oval or roundish nuclei, without any significant pleomorphism. The architectural pattern of the epithelial/myoepithelial component was tubular, glandular, and cribriform, in cords, islets, and groups. The stroma was fibrous, hyalinized with sparse psammoma bodies (H&E stain, 20×)



the neoplasm, the differential diagnosis should be made accordingly by an experienced pathologist.

Even the evidence of atypia or vascular invasion can be seen, mimicking malignant transformation, but these features may still be acceptable for adenomas under the appropriate histological settings. Bone involvement is not considered a typical finding. A translocation between genes *PLAG1* and *HMG2* is associated with the incidence of pleomorphic adenomas [7]. This is a possible diagnostic key examination for difficult cases.

As of any pleomorphic adenoma, a complete excision of the lesion is the standard of care in suspected pleomorphic adenomas of the palate. High recurrence is recorded in case of enucleation of the lesion or dissection of the capsule and tumour spillage intraoperatively. Bone removal is strongly recommended if the bone is infiltrated. Since malignant transformation of PAs (carcinoma ex pleomorphic adenoma) is recorded in an incidence of 6%, it is of utmost importance to safely excise the entire lesion [8].

Concluding, pleomorphic adenomas of the palatal minor salivary glands remain a rather rare clinical condition, and it is unusual to infiltrate the adjacent palatal bone. A thorough clinical imaging and histological/cytological examination should always be performed to define the benign or malignant nature of the lesion, preoperatively. Adequate excision with adequate margins and in toto excision of the tumour is important for minimizing the recurrence rate and malignant transformation. Long-term follow-up is recommended.

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Declarations

Ethics Approval Not applicable.

Consent to Participate A written consent was obtained by the patient.

Consent for Publication A written consent was obtained by the patient for publication.

Conflict of Interest The authors declare no competing interests.

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