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Case Report

Pleomorphic Adenoma Mimicking Periapical Cyst: Case Report

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Abstract:

Pleomorphic adenoma is the most common salivary gland neoplasm. The lesion presents as a firm, slow-growing, painless mass. It can occur at any age, but is most common in young adults between the ages of 30 and 50 years. The diversity of the morphological histopathological pattern is one of its most characteristic features. The purpose of this article is to report a case of pleomorphic adenoma on the palatal mucosa of a 15-year-old patient, mimicking a periapical cyst. We performed an incisional biopsy of the lesion and diagnosed the pleomorphic adenoma, and then performed a total excision of the lesion. The diagnostic and therapeutic features of pleomorphic adenoma were discussed.

Keywords: *pleomorphic adenoma; salivary gland neoplasms; minor salivary glands; oral surgery; oral diagnosis.*

Introduction

The pleomorphic adenoma, or mixed benign tumor, is the most common benign salivary gland neoplasm, accounting for 40% to 70% of all tumors of these structures. It occurs most frequently in the parotid gland, followed by the submandibular gland and minor salivary glands. The most common site of pleomorphic adenoma of minor salivary glands is the palate, followed by the upper lip, jugal mucosa, floor of mouth, tongue, tonsil, pharynx, retromolar area and nasal cavity[1-8].

The lesion manifests as a firm tumor mass; fibroelastic in consistency; slow growing and painless; covered by normal or sometimes ulcerated mucosa; with 10 to 20 mm in diameter. When pleomorphic adenoma affects the palatal mucosa, it can make chewing, swallowing, and phonation difficult[1,3-5]. If neglected, it can reach larger proportions[1]. Eventually, the parotid lesion can grow medially between the ascending ramus and the stylomandibular ligament, resulting in a tumor that presents as a mass of the lateral pharyngeal wall or soft palate[7,8].

It can occur at any age, being more common in young adults between the ages of 30 and 60 years, and greatest predilection for the female gender[1,2].

From the histopathological point of view, it may present epithelial, myoepithelial, ductal and stromal cells and may contain areas with squamous keratinizing cells, myxoid, fibrous, cartilaginous and bony areas intermingled in a background similar to mesenchyme[1,2,4,6,9]. Its malignant transformation, resulting in a carcinoma ex pleomorphic adenoma is cited to occur in about 3 to 6% of cases[4,10,11].

The treatment for pleomorphic adenoma of the palate consists of local surgical excision including its covering mucosa, with normal tissue, in order to avoid recurrence. Pleomorphic adenoma has a tendency to recur if not completely excised, ranging from 2 to 22%[2,11,12]. The purpose of this article is to report a case of pleomorphic adenoma on the palatal mucosa of a 15-year-old patient, mimicking a periapical cyst.

Case Report

A Caucasian female patient, 15-year-old, presented to the Brazilian Air Force Hospital in São Paulo, complaining of a lesion on the palate.

Clinically, a nodule was observed on the left palatal mucosa, in the periapical region of teeth 23, 24 and 25. The lesion was asymptomatic; the mucosa was normal in color; the surface was intact, without ulcerations; it had a fibroelastic consistency; the base was sessile; it was approximately 12mm in diameter; and it had evolved for 2 years (Figure 1).

The initial panoramic radiograph showed a radiolucent image circumscribed by a radiopaque halo in the periapical region of teeth 23, 24 and 25 (Figure 2). Later and complementarily, after the start of orthodontic treatment, the images were observed on periapical and occlusal radiographs (Figures 3 and 4, respectively). The radiographic images directed the diagnostic hypothesis to periapical cyst.

Figure 1: *Lesion on the left palatal mucosa, in the periapical region of teeth 23, 24 and 25.*

Figure 2: *Radiolucent image circumscribed by a radiopaque halo in the periapical region of teeth 23, 24 and 25.*

Figure 3: *Radiolucent image on periapical radiograph.*

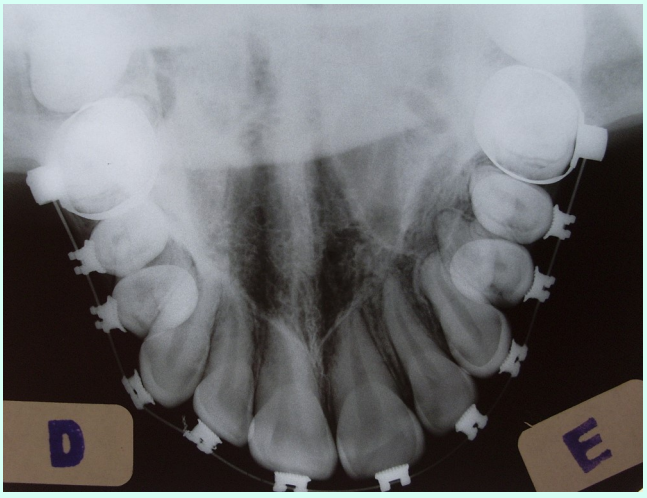

Figure 4: *Radiolucent image on occlusal radiograph.*

Incisional biopsy was recommended prior to the surgical procedure. The information was forwarded to the adolescent's parents, and after all doubts were clarified, the surgical procedure was scheduled.

Under local infiltrative anesthesia, an incision was made around the lesion and removed at its base (Figures 5 and 6, respectively). The region was sutured. The patient was prescribed analgesic medication.

The removed fragment (Figure 7) was fixed in 10% formalin and sent to the Laboratory of Surgical Pathology of the Department of Oral Pathology of the School of Dentistry, University of São Paulo. The histopathological examination revealed a mixture of epithelial, glandular, myoepithelial, and mesenchymal stromal components with areas of myxoid degeneration. The epithelial component showed areas of squamous metaplasia. Next to the myoepithelial component hyalinization of the stroma was noted. No signs of malignancy such as foci of capsular, vascular or perineural invasion were observed (Figure 8). The final diagnosis was pleomorphic adenoma.

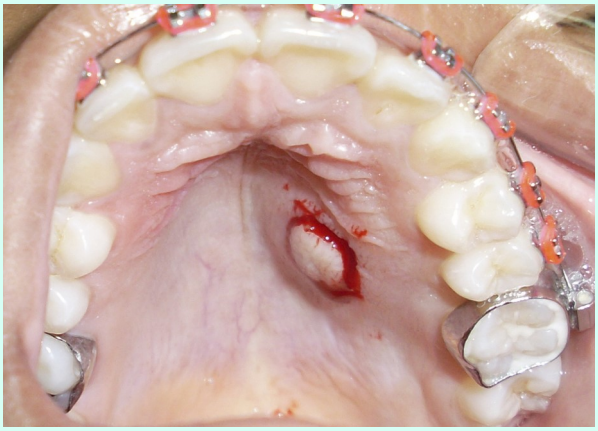

Figure 5: *Incision was made around the lesion.*

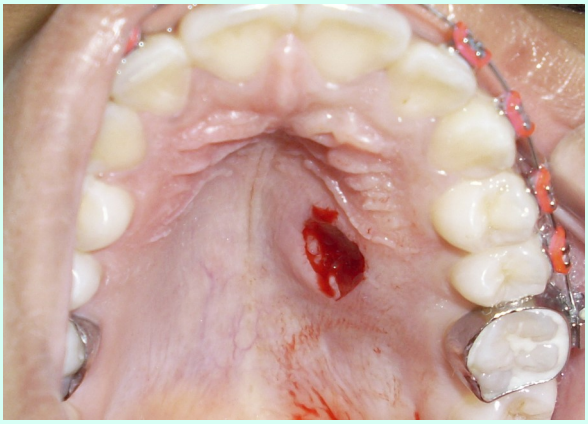

Figure 6: *Surgical site after removal of the lesion*

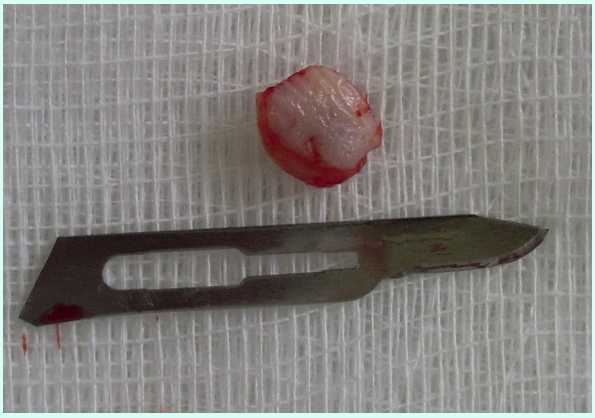

Figure 7: *Lesion removed, after incisional biopsy*

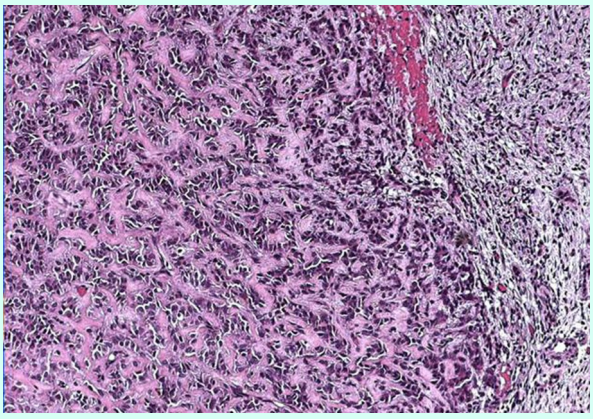

Figure 8: *Histopathological aspects of the pleomorphic adenoma (magnification: 40X)*

After 15 days, the remaining sutures were removed. No complaints or complications were reported.

After 30 days, with complete repair (Figure 9), exeresis of the lesion was performed. Under local anesthesia on the palatal mucosa, an intrasulcular palatal incision was made on the left side, between teeth 13 and 26. The gingival flap was folded back, exposing the lesion (Figure 10). The lesion was enucleated (Figure 11) and the bone cavity was cleaned and washed thoroughly with cooled saline solution (Figure 12). The region was sutured. The patient was prescribed analgesic, anti-inflammatory and antibiotic drugs. The removed lesion (Figure 13) was referred back to the Surgical Pathology Laboratory and the final diagnosis was pleomorphic adenoma.

After 10 days, the remaining sutures were removed. No complaints and or complications were reported. The patient was evaluated after 40 days, with satisfactory total healing (Figure 14).

After 3 months, a new clinical (Figure 15) and radiographic (Figures 16 and 17) evaluation was performed. The patient has been followed up for 2 years with no signs of recurrence.

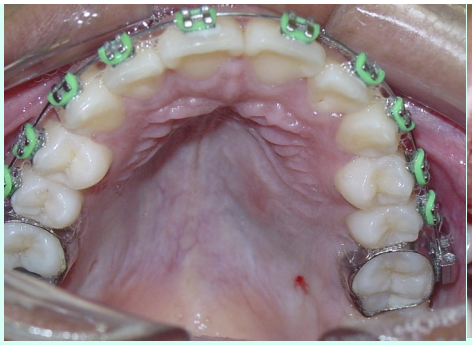

Figure 9: *Complete healing (after 30 days of the incisional biopsy)*

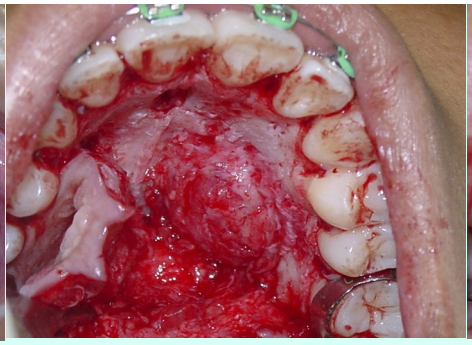

Figure 10: *Intrasulcular palatal incision, with folded gingival flap*

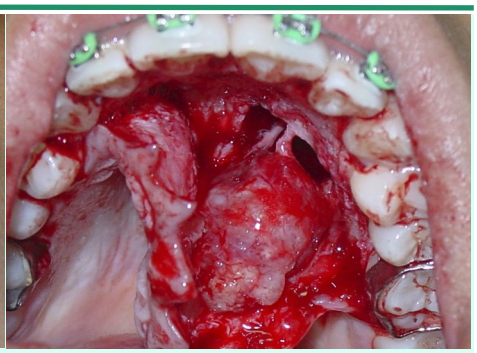

Figure 11: *Enucleation of the lesion*

Figure 12: *Cleaning of the bone cavity*

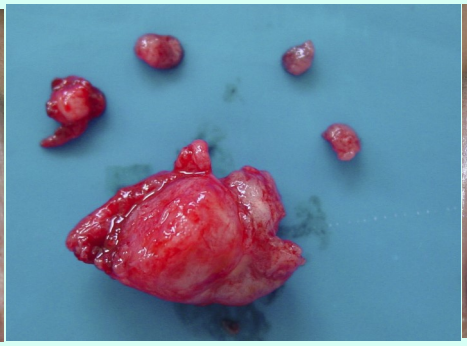

Figure 13: *Lesion completely*

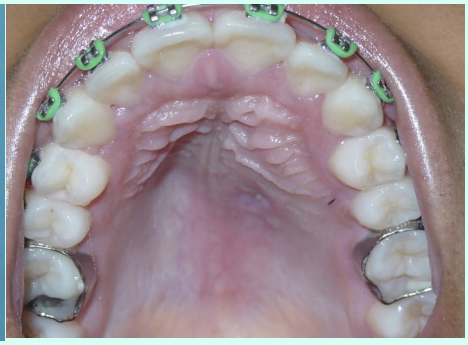

Figure 14: *Satisfactory total healing* (after 40 days)

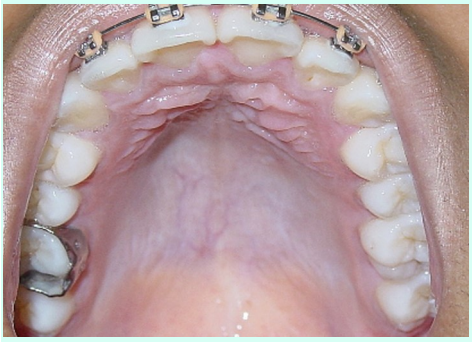

Figure 15: *Clinical evaluation (after 3 months)*

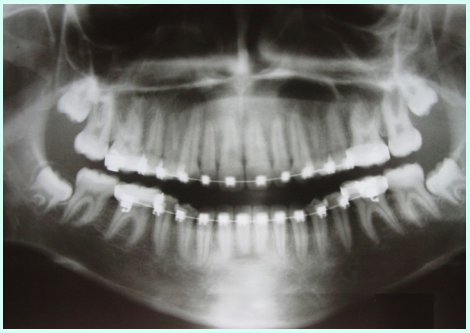

Figure 16: *Radiographic evaluation (after 3 months): panoramic radiograph*

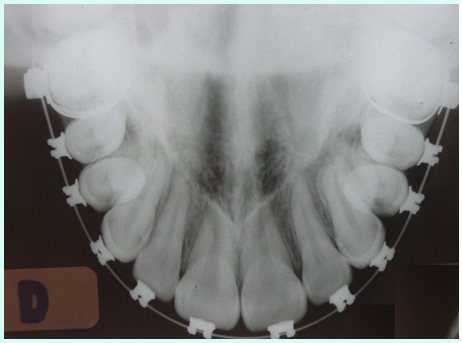

Figure 17: *Radiographic evaluation (after 3 months): occlusal radiograph*

Discussion

Pleomorphic adenoma is a benign neoplasm that occurs most frequently in larger salivary glands. It accounts for approximately 70% of all reported benign salivary gland tumors[4,8,13]. About 75% to 85% of pleomorphic adenomas are located in the parotid, 5 to 10% are located in the submandibular gland, and 10% in the minor salivary glands[14]. Of these, the most affected are those of the palate, upper lip, jugal mucosa, floor of mouth, larynx and trachea[1,2,4,8]. As in the case presented here, the most prevalent site of pleomorphic adenoma in the palatal region is the posterolateral area of the hard palate.

When located in the soft palate it can reach large proportions, causing difficulties in chewing, swallowing, speaking and breathing[6-8]. In this case, the lesion was not large enough to cause significant functional disorders, causing only mild discomfort when the patient swallowed. Although the pleomorphic adenoma may affect patients at any age, it mainly affects individuals from the 4th to 6th decades of life, and is rare in children[2,9]. In the present report, the patient was an adolescent (15 years-old).

The presumptive clinical and radiographic diagnosis of this case was periapical cyst. However, it was only elucidated by incisional biopsy, whose histopathological examination revealed it to be a pleomorphic adenoma. Complementary exams, such as fine needle aspiration puncture or previous incisional biopsy are the exams performed to define the diagnosis[2,4,8]. Inside the mouth, due to the practicality and absence of complications, when compared to extra-oral accesses to major salivary glands, incisional biopsy is still the most used method to reach the definitive diagnosis[8].

Importantly, the differential diagnosis of lesions found on the palate includes malignant neoplasms of minor salivary glands such as mucoepidermoid carcinoma, low-grade polymorphous adenocarcinoma, and adenoid cystic carcinoma. Mesenchymal lesions such as neurofibroma and rhabdomyosarcoma can also present as palatal volumetric enlargement[9].

The recommended treatment is exeresis of the lesion with the covering mucosa, with the purpose of avoiding recurrence of the lesion[1,2,4,9,12]. In the present case, the mucosa was removed already in the first approach (incisional biopsy).

Malignant transformation may occur in 3% to 6% of cases of pleomorphic adenoma, resulting in carcinoma ex pleomorphic adenoma[4,10,11,15]. This possibility, even if reduced, determines that the clinical and radiographic post-surgical follow-up should be performed in the long term[1].

Conclusion

Pleomorphic adenoma is the most common salivary gland neoplasm, occurring most frequently in the parotid gland, followed by the submandibular gland and minor salivary glands. The recommended treatment is total exeresis of the lesion. However, for the elucidation of the diagnostic hypothesis, it is recommended that an incisional biopsy be performed prior to the surgical approach. In view of the possibility of recurrence of the lesion, strict clinical and radiographic postoperative control should be performed.

Conflict of Interest

The authors declare no conflict of interest.

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