

PLEOMORPHIC ADENOMA: CLINICAL CASE REPORT

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Abstract: Pleomorphic adenoma is a mixed, benign tumor with more common occurrence in the salivary glands, mainly in the parotid and submandibular gland, being less frequent in the intraoral region, when it occurs, it has a preference for the palate, lip and buccal mucosa. The objective of this work is to present two clinical cases of Pleomorphic Adenoma with different and atypical locations, and to choose the best form of treatment for each specific case, as well as to describe the clinical stages of management with the patient from surgery to the postoperative period. The present research took place in an HEI with two patients diagnosed with pleomorphic adenoma, one in the hard palate and the other in the upper lip. The diagnosis was made through biopsy and histopathological examination. In view of the above, we can observe that the Pleomorphic Adenoma is a pathology that deserves to be highlighted, given that it has particular characteristics, and the knowledge of the Dental Surgeon about this pathology is extremely important, in order to carry out an early diagnosis. These peculiar characteristics will help in the most specific conduct for each region, always aiming to reduce the chances of malignancy and recurrence.

Keywords: Pleomorphic adenoma. Pathology. Surgery. Palate. Upper lip.

INTRODUCTION

According to Neville *et al.* (2016), Pleomorphic Adenoma (PA), also known as benign mixed tumor, is the most common salivary gland neoplasm and occurs mainly in the parotid and submandibular glands. Adenomas are derived from a mixture of ductal and myoepithelial elements, and there may be great microscopic diversity from one neoplasm to another, as well as in different areas of the same tumor.

Most are asymptomatic, with a firm consistency and slow growth. It is common

to have an ovoid conformation and well-defined margins, and may present mobility, except in the palate, in which they are found in the posterolateral region at the junction of the hard and soft palate and the size varies from millimeters to centimeters. It frequently affects females between the ages of 30 and 60 years. (BIGUELINE *et al.*, 2015; SILVA *et al.*, 2008; SINGH *et al.*, 2019)

Treatment is crucial because of the potential for long-standing lesions to undergo malignant transformation. Most of the management is surgical, and complete excision of the lesion is essential to avoid possible recurrence. All aspects must be observed in detail and the characteristics of the present lesion must be compared with the existing data, this way the conduct will be specific for each region, whether in the upper lip or hard palate (MAIR; AGUIRRE, 2016; SILVA *et al.*, 2008). With proper surgical removal, the prognosis is excellent, with a cure rate of more than 95 %, and the risk of recurrence is lower for pleomorphic adenomas in minor salivary glands (NEVILLE *et al.*, 2016).

The objective of the present work is to present two clinical cases of Pleomorphic Adenoma with different and atypical locations, and to choose the best form of treatment for each specific case. It is of great importance in the literature and there are few cases recorded because they are in atypical regions, in addition to the patients having unusual particularities to those recorded in the literature.

THEORETICAL FRAMEWORKS

According to Estevão *et al.* (2015) the salivary glands are a set of glands with serous, mucous or seromucous characteristics. It may present benign pathologies, with different forms of treatment. Among the most frequent pathologies, we highlight sialolithiasis and benign tumors of the salivary glands, among them pleomorphic adenoma.

For Neville *et al.* (2016) pleomorphic adenoma, or benign mixed tumor, is the most common neoplasm in salivary glands. This lesion represents about 50 % to 77 % of parotid neoplasms, 53 % to 72 % of submandibular gland tumors, and 33 % to 41 % of minor salivary gland tumors and are derived from a mixture of ductal and myoepithelial elements. They present, microscopically, cellular diversity, whether between areas of the same tumor or in different neoplasms. The adenoma has histopathological variations, so that its characteristics do not follow a parameter of the benign tumors seen in the literature. Although the cellular pattern is variable, individual cells rarely have variations in shape, but even then, it is called a mixed tumor.

According to Das Neves *et al.* (2007) salivary gland neoplasms are rare and their pathogenesis is uncertain as well as the predilection for oncogenesis. According to Israel *et al.* (2016) this is due to its complexity also attributed to the heterogeneity of the cells of origin of these lesions. Pathological diagnosis is the key to the proper management of these lesions, since the degree of aggressiveness depends on their histological types.

For Oliveira *et al.* (2016) and Bigeline *et al.* (2015) the lesions of the pathology, when intraoral, occur preferentially on the palate followed by the lip and cheek mucosa, as reported in the present work. It can affect individuals in any age group, especially in the third and sixth decade of life, however this work differs in terms of the common age found. Agreeing with the predominance of the female gender (approximately 60%), but without justification for such a predilection.

A very common region is the palate, which stands out in the literature with a percentage of approximately 50 % to 65 % of intraoral examples. This location is followed by the upper lip (19 % to 27 %) and the buccal mucosa (13 % to 17 %). It is strongly adhered

to the adenoma, and, therefore, the neoplasms of this location are not mobile, being mostly surrounded by a circumscribed capsule, unlike the labial mucosa or the buccal mucosa, which are mobile.

Regarding the incidence in the lip, we can see a higher incidence in the upper lip, as it is a region with a higher rate of anomalies, when some variation is found, several pathologies are suggested, but the 2nd most common pathology in the upper lip is the Pleomorphic Adenoma, even being rare in this location, the best way to classify is by clinical examination, through palpation of submucosal swellings, to differentiate from cysts and tumors, accompanied by histopathological study to obtain a more accurate and definitive diagnosis (SINGH *et al.*, 2019).

According to Neville *et al.* (2016) and Queiroz *et al.* (2014), the clinical features are nodular lesions with a smooth surface and a firm, painless, slow-growing swelling that does not attach to adjacent tissue (mobile lesion except on the hard palate).

Histologically, Pleomorphic Adenoma it is a normally encapsulated and well-circumscribed lesion (FIGURE 1), which may also present an incomplete capsule and exhibit infiltration by neoplastic cells. Myoepithelial cells usually comprise a large percentage of neoplastic cells and exhibit variable morphology, sometimes appearing as angled or spindle-shaped cells. Some myoepithelial cells are rounded and have an eccentric nucleus and hyalinized eosinophilic cytoplasm, resembling plasma cells (FIGURE 2). These characteristic plasmacytoid myoepithelial cells are most prominent in neoplasms arising from minor salivary glands. Microscopically, some fat cells or osteoid material can also be observed (NEVILLE *et al.*, 2016).

The diagnosis is generally made based on a detailed clinical history and with the aid of exams. Imaging exams, although not essential,

have shown an important role in establishing the origin, location and limits of the lesion, especially CT scans.

As for the biopsy of the lesion, it must always be considered, both to exclude other possible malignant lesions and to assess whether the lesion has a chance of becoming malignant. It is recommended that a surgical procedure be performed, so that surgical excision is the best way to access the lesion, it must be wide with free margins, with the intention of further reducing the rate of recurrence, as well as avoiding transformations. evil. The possibility of recurrence occurs when the nodule is not completely removed, it is also worth remembering that, even with the total removal of the nodule, clinical surveillance is essential for early diagnosis in cases of recurrence or malignancy, and follow-up after surgery is essential (MELO *et al.*, 2016; ROCHA *et al.*, 2015).

With proper surgical removal, the cure rate is 95% and has an excellent prognosis. The risk of recurrence is lower for pleomorphic adenomas of the minor salivary gland. The choice of surgical technique must be related to the location, total size of the nodule and better patient comfort.

It is also important to remember that after the removal surgery, follow-up must be carried out every year to observe possible changes and maintain control of non-recurrence (CARVALHO *et al.*, 2017; SENGUL; SENGUL, 2011).

The likely risk of malignant transformation is small, but it may occur in approximately 3 % to 4 % of all cases. The risk increases with the time of evolution of the neoplasm (DAS NEVES *et al.*, 2007; NEVILLE *et al.*, 2016; SENGUL; SENGUL, 2011). The potential for malignant transformation of Pleomorphic Adenoma has been observed, mainly in cases of incomplete surgical excision.

The risk of local recurrence of Pleomorphic

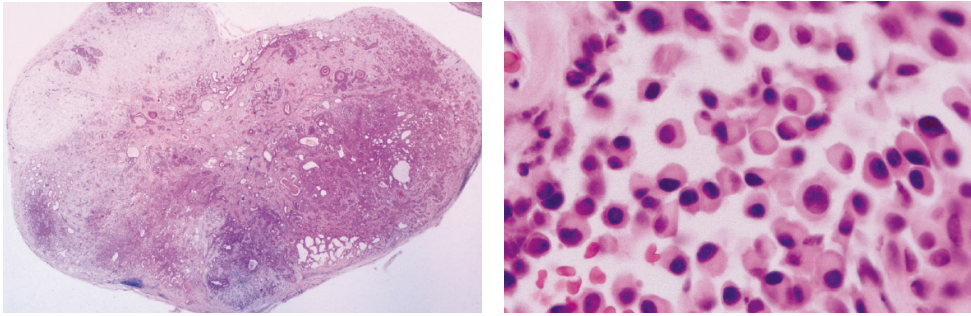


FIGURE 1: Capsule that surrounds the Pleomorphic Adenoma and can be seen even at the lowest magnification. FIGURE 2: Myoepithelial cells.

Source: NEVILLE *et al.* (2016)



FIGURE 3: Initial clinical examination, highlighting the superior region of the mucosa, in which a bulge can be observed under the region of the lateral incisor and the canine of the direct hemi-arch

Source: BORBA; SOARES (2020)



FIGURE 4: Incision at the bottom of the sulcus, extension from the lateral incisor to the maxillary first premolar of the right hemi-arch. FIGURE 5: Removal of the specimen with the aid of dissection forceps and the Minissota retractor

Source: BORBA; SOARES (2020)

Adenoma after the first surgery is associated with the presence of clinicopathological variables that include: age less than 30 years since diagnosis, presence of extracapsular neoplasm in the form of satellite nodules, variant rich in stroma and incomplete surgical excision; the time of evolution of the neoplasm since the diagnosis, probably, results from the accumulation of genetic alterations.

METHODOLOGY

This study complied with Resolution No. 466/2012 of the National Health Council (CNS) and was submitted to the Research Ethics Committee – CEP of the institution: “Centro Universitário Integral Diferencial” (UNIFACID) via Plataforma Brasil. Data collection started only after approval of the Project by the CEP- (Appendix A).

The research is characterized as a study of an applied nature, with a qualitative approach, with exploratory objectives and a case study type.

It took place in a Higher Education Institution (HEI) in the city of Teresina-PI, in a dental school clinic.

Two patients participated in the study, diagnosed with Pleomorphic Adenoma in the upper lip and the other in the hard palate.

Inclusion criteria: patients who have a Pleomorphic Adenoma in the upper lip and another in the hard palate confirmed by histopathological examination were included in the sample.

The case study consists of reporting the clinical steps and the management of the Dental Surgeon with the patient with pleomorphic adenoma, from the first consultation to the referral and result of the histopathological examination, as well as the postoperative period and long-term follow-up. term, since in cases that have recurrence, the diagnosis will be earlier.

About the risks: the participants of this

study were exposed to risks from the surgical intervention, accidents/complications/consequences that may result from the biopsy such as: hemorrhage, postoperative pain and infection, which were minimized through the best surgical technique for the case., preserving the aseptic chain and making the appropriate drug prescription. In its benefits: patients in addition to having received the benefit of the diagnosis of the pathology (biopsy + histopathological examination), in addition to all dental follow-up after the end of the research. Having still an indirect benefit, where the results of this research characterize cases with a low incidence, this will contribute to help the literature to better elucidate the characteristics of this lesion that still has undefined pathogenesis.

DATA COLLECTION

Data collection started after receiving approval from the Research Ethics Committee. The present study selected two patients diagnosed with Pleomorphic Adenoma. The diagnosis was made through biopsy and histopathological examination. Postoperative follow-up was carried out until the healing of the surgical wound and six months after the surgery, continuing with the follow-up as the patient’s age progressed. All steps were recorded and duly documented.

DATA ORGANIZATION AND ANALYSIS

Data were analyzed by clinical records, through questions and photographs, histological slides were made for analysis and their reports were verified. The clinical steps were described, from the first consultation to the biopsy result and the procedure for total removal of the benign tumor.

CLINICAL CASE REPORTS

CLINICAL CASE

Pleomorphic adenoma in upper lip

Patient CCS, female, 24 years old, black, attended the clinic of the Faculty of Dentistry, UniFacid, located in the city of Teresina, Piauí. On clinical examination (FIGURE 3) a nodule was observed in the region of the vestibule fundus and upper lip on the right side, with normal color, without painful symptoms, measuring more or less 10 mm. The patient has no history of allergic reactions to anesthesia, and reported that the nodule has evolved over the past 3 years.

No radiographic examination was requested, given that it was in soft tissue. The conduct of choice was to request an anatomopathological examination through excisional biopsy. The specimen was fixed in 10% buffered formalin and sent for analysis with the diagnostic hypotheses of Gland Tumor or Pleomorphic Adenoma.

The surgery was performed following all the precepts of the aseptic chain, seeking the best comfort for the patient and the surgeon. Excisional biopsy was the final choice because the affected region causes discomfort to the patient, in addition to the lesion having small diameters.

The incision was performed at the bottom of the sulcus, extending from the lateral incisor to the maxillary second premolar of the right hemi-arch (FIGURE 4). Removal of the specimen required the aid of dissection forceps and the Minissota retractor (FIGURES 5). The minor salivary glands were preserved (FIGURE 6) and immediately after a thorough check the flap was closed with simple stitches (FIGURE 7). The specimen was sent to the biopsy center (FIGURE 8) and after 15 days we obtained the final report.

Given the analysis of the report, the suspicion of Pleomorphic Adenoma was confirmed and the fragment was detailed with

macroscopic and microscopic characteristics.

Macroscopically, it was possible to observe a fragment with a globular shape, irregular surface, fibrous consistency and whitish color, measuring 13mm x 11mm x 10mm.

In histological sections, microscopically, the neoplasm fragment is characterized by proliferation of epithelial and myoepithelial cells organized in a pattern, predominantly ductiform regions, an inner layer composed of eosinophilic cells and an external layer composed of neoplastic cells. They have rounded, fusiform, epithelioid and plasmacytoid morphology, and there is also a region with dense hyalinized connective tissue surrounding the lesion, characterizing the neoplasm of glandular origin.

The patient was informed about the result and after 6 months a consultation was carried out for control and observation of the lesion site (FIGURE 9), given the clinical characteristics, it was observed that there was no sign of recurrence and the mucosa was healed and in perfect condition.

CLINICAL CASE

Pleomorphic Adenoma in Hard Palate

Patient VNS, female, 16 years old, mixed race, with Rheumatoid Arthritis, but without dysfunctional manifestations, it was a young and healthy patient. He attended the dental school clinic of an HEI, located in the city of Teresina, Piauí, in search of dental treatment, reporting the presence of a nodule in the "roof of the mouth". Regarding clinical examination, it was possible to observe the presence of the nodule in the posterolateral region of the hard palate, close to the posterior teeth on the left side (FIGURE 10), without painful symptoms, with normal color, measuring approximately 30 mm, with no history of allergy to anesthesia. The same reported that since the appearance to the current size had already passed more or less two years.



FIGURE 6: Mucous tissue pinched with Addison 's forceps, the preserved minor salivary glands can be observed. FIGURE 7: Closing the flap with simple stitches

Source: BORBA; SOARES (2020)

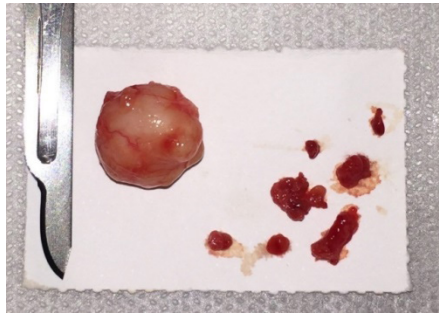


FIGURE 8: Lesion after complete removal, containing some fragments for safety margins.

Source: BORBA; SOARES (2020)



FIGURE 9: Postoperative period of the region after 6 months.

Source: BORBA; SOARES (2020)

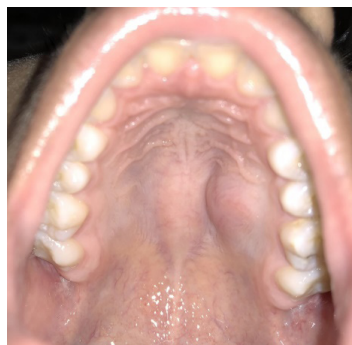


FIGURE 10: Clinical examination showing the presence of the nodule in the posterolateral region of the hard palate, close to the posterior teeth on the left side.

Source: BORBA; SOARES (2020)

As it is a region of the maxilla and is in soft tissue, no additional examination was necessary, given that because there are several structures in the maxilla, the examination could have overlapped, not being effective for full visualization of the nodule. The conduct of choice was to request an anatomopathological examination through the surgical procedure of total removal of the nodule, excisional biopsy. The specimen was fixed in 10% buffered formalin and sent for analysis with the diagnostic hypothesis of Pleomorphic Adenoma.

The surgery was performed following all the precepts of the aseptic chain, seeking the best comfort for the patient and the surgeon. Excisional biopsy was the final choice due to the affected region causing discomfort to the patient.

The incision was made through the palate, delimiting the site of the nodule, comprising the premolars and molars region of the left hemi-arch, below the periosteum and prominent mucosa (FIGURE 11). It was necessary to dissect the tissues well to gain access to the nodule and remove the specimen, for which the Molt detacher and the Lucas curette were used (FIGURE 12).

Removal was performed with Allis forceps, removing the lesion completely (FIGURE 13), leaving only the capsule. After total removal of the fragment, the capsule was removed with a Molt detacher, which involved it (FIGURE 14), and later, to control the blood, the region was clamped with a hemostatic forceps.

After the capsule was completely removed, an inspection was carried out to see if there were any more fragments, also performing the curettage of possible fragments that still existed (FIGURE 15). After a thorough check, gauze was used to contain the bleeding, with stable bleeding, the flap could be closed. The suture was performed with simple stitches (FIGURE 16) and the specimen was sent to

the biopsy center, after one month we obtained the final report.

Receipt of the report, the result was positive for Pleomorphic Adenoma in the hard palate. In its macroscopic specifications, it was observed the presence of a nodular fragment measuring about 15mm x 10mm x 07mm, with a smooth surface, rubbery consistency and whitish color.

On the other hand, the characteristics of the histological sections reveal fragments of epithelial neoplasia of glandular origin. Having two types of cells, luminal cells and myoepithelial cells, predominantly myoepithelial cells. Arranged in the form of blocks, trabeculae and sheet, exhibiting predominantly plasmacytoid morphology.

The patient was informed of the result and the suture was removed. After 6 months, a consultation was carried out to control and observe the lesion site (FIGURE 17), given the clinical characteristics, it was observed that there was no sign of recurrence and the mucosa was healed and in perfect condition.

DISCUSSIONS

Pleomorphic adenoma is the most common neoplasm of the salivary glands, the term pleomorphic results from the histopathological diversity of the tumor, with variability of characteristics from one neoplasm to another or in the same neoplasm. The essential components are the capsules, with epithelial and myoepithelial cells, also presenting mesenchymal elements, the same epithelium can also form ducts and cystic structures, in addition to eosinophilic cells and hyalinized cytoplasm (ERDEM *et al.*, 2011; NEVILLE *et al.*, 2016); QUEIROZ *et al.*, 2014; TAIWO *et al.*, 2018).

The two cases reported in this study corroborate all the characteristics mentioned above, given the observation that the presence of capsules can be seen in cases that do not



FIGURE 11: Incision in the periosteum and mucosa, premolars and molars region. FIGURE 12: Divulsion with Molt 's peeler and Lucas' curette.

Source: BORBA; SOARES (2020)



FIGURE 13: Nodule was clamped with Allis forceps, removing the lesion completely. FIGURE 14: R Capsule Emotion, with Molt 's Peeler

Source: BORBA; SOARES (2020)



FIGURE 15: Inspection and curettage of any fragment not removed. FIGURE 16: Simple stitch suture.

Source: BORBA; SOARES (2020)



FIGURE 17: Postoperative period of the region after 6 months.

Source: BORBA; SOARES (2020)

have the presence of neoplastic cells in the structure, given that Neville *et al.* (2016) observed in their studies that in lesions that had neoplastic cells, the capsule was found incomplete because these cells infiltrated the capsule produced. When we followed the cells present in the histopathological examination of the two cases addressed in the study, we observed that the neoplasm located in the upper lip has neoplastic cells and an incomplete capsule. In the case of neoplasms found in the hard palate, the capsule is present, leading to the conclusion that the nodule is more common in the hard palate region, as also highlighted by studies carried out by Oliveira *et al.* (2016) and Rahnama *et al.* (2013).

Regarding the prevalence of age and gender, the literature records the highest incidence in females, which is confirmed in both case reports. Regarding age, it is interesting to note that both patients in the study do not reinforce the statistics found by Neville *et al.* (2016); Oliveira *et al.* (2016); Singh *et al.* (2019); taiwo *et al.* (2018), for example, who present more common cases in the age group of 30 to 60 years. However, in the studies by Carvalho *et al.* (2017) and Queiroz *et al.* (2014) the authors register cases where the exposed patient is also younger than the common age, as seen in this study, showing the importance of early diagnosis, knowledge of the general dentist and the corrector referral to the specialist dentist. If the correct removal of existing cells in the adenoma is not carried out, certain cells may proliferate again over time, which may increase the chance of recurrence.

The treatment of choice for the pleomorphic adenomas exposed in the present study was surgical excision of the nodule, which is defined by the literature as the most effective approach, with a good prognosis, authors such as Carvalho *et al.* (2016); Estevão *et al.* (2015); May; Aguirre (2016) and Silva *et al.*

(2008) performed similar conduct.

With adequate surgical removal, the prognosis is excellent, with a cure rate of more than 95%, and recurrence in smaller tumors of the salivary gland is rare, but the possibility still exists, and the diagnosis by a trained professional is of paramount importance and the long-term follow-up (NEVILLE *et al.*, 2016; SINGH *et al.*, 2019; TAIWO *et al.*, 2017). In the study described, the initial follow-up was carried out six months after the surgery and, over time, the patient will follow up with his dentist for observation and control of the regions.

CONCLUSIONS

Based on case reports and literature review, we can conclude that:

The clinical and histological characteristics and its evolution are similar to those found in the literature;

Total removal of the lesions, by surgical excision, was the best approach;

Patient discomfort was reduced through a conservative incision, bleeding containment, and firm suturing. It is necessary to follow up after the treatment is finished.

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