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PLEOMORPHIC ADENOMA IN THE MINOR SALIVARY GLAND - CASE REPORT

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Abstract: Pleomorphic adenoma, also known as benign mixed cell tumor, is composed of mixed cells, due to the combination of myopithelial and ductal elements. The lesion presents unusual histopathological characteristics, as none of them is absolutely specific. The aim of this study is to review the literature on Pleomorphic Adenoma of the minor salivary gland and to present a clinical case of a patient affected by Pleomorphic Adenoma of the minor salivary gland. An integrative literature review was carried out, based on electronic databases: *Medline* via *Pubmed, Scielo* and *LILACS*. Publications in Portuguese, Spanish and English that addressed concepts relevant to the topic were included. A 38-year-old female patient sought care to evaluate an increase in intraoral volume in the palate region with a 3-year evolution. The proposed treatment was removal of the lesion, with safety margins, under general anesthesia, due to the posterior location of the neoplasm. Therefore, it is emphasized that the correct diagnosis, choice of histopathological analysis, good quality radiographic examinations and well-indicated surgical technique promote a better prognosis for the patient with a low probability of recurrence.
Keywords: Minor Salivary Gland Pleomorphic Adenoma; Pleomorphic adenoma; Salivary Gland Tumor.

INTRODUCTION

Tumors in the salivary glands are rare among tumors that affect the head and neck region, representing 3 to 6%, the most affected region of the glands is the parotid region and the most common is the pleomorphic adenoma (SILVA, 2020). Neoplasms can affect both the minor and major salivary glands. The same, are characterized in the same way, differentiating the location of the tumor and extension of the lesion (SANTOS, 2021).

According to studies by organizations such as the World Health Organization (WHO), approximately 30 classifications of neoplastic lesions involving the salivary gland are recognized. About 2 to 6% of tumors in the head and neck region correspond to those involving the salivary glands (SANTOS, 2021).

Pleomorphic adenoma, also known as benign mixed cell tumor, is composed of mixed cells, due to the combination of myopithelial and ductal elements. The present lesion has unusual histopathological characteristics, as none of them is absolutely specific (NEVILLE, 2011). Cell plurality can exist in different areas of the tumor and not just from one tumor to another, thus giving the characteristic of a mixed tumor (GAMA, 2018).

According to the cystic extension of the lesion, in histopathological examinations, they may exhibit particularities, such as in their patterns, which may be loose, chondroid, collagenous or hyalinized (SANTOS, 2021). In clinical examinations, some characteristics resemble those of other lesions, which may influence the diagnosis, requiring confirmation of the diagnostic hypothesis (SANTOS, 2021).

Regarding its location, about 50% to 77% of the cases occur mainly the parotid glands, the involvement of the submandibular gland occurs in 53% to 72%, and in the range of 33% to 41% it affects the minor salivary glands, the which makes the region rarer (GAMA, 2018).

The size of the gland involved will associate the neoplasm, it is not uncommon that tumors that reach the greater salivary gland have a greater chance of being benign, and those that reach the smaller one, predispose it to be malignant (DE SOUSA, 2019).

Neoplastic lesions of the salivary glands have lower incidence rates of malignancy, with diagnoses of benign tumors prevailing (DE SOUSA, 2019).

The usual area of pleomorphic adenoma of the minor salivary gland is the region of the palate, subsequent to the area of the upper lip and jugal mucosa, floor of the mouth, retromolar area, nasal cavity, tonsil and pharynx (SANTOS, 2016).

Pleomorphic adenoma has a predilection in women of any age group, with a prevalence between the 4th and 6th decade of life (GAMA, 2018). It is clinically characterized by presenting a firm nodule, mobile to palpation, well-defined, painless and slow-growing. Less frequently, it can exhibit ulceration and rapid growth (RODRIGUES, 2018). The differential diagnosis is made through histopathological examination, due to the instability of the parenchyma and stroma in the diverse morphological pattern of the lesion (NEVILLE, 2016).

The treatment for pleomorphic adenoma in the minor salivary gland consists of enucleation of the lesion, which will obtain a satisfactory prognosis in terms of recovery and low recurrence (MAIA, 2019).

The recurrence of the lesion occurs in the range of 0.4 to 45%, and malignancy may occur with recurrence. With this, the correct diagnosis and treatment plan is extremely important, to avoid relapses and worsening of the patient's quality of life (SANTOS, 2021).

The objective of the present work is to carry out a review of the literature on Pleomorphic Adenoma of the minor salivary gland, showing its characteristics: clinical, radiographic and histopathological aspects; diagnosis, treatment and prognosis. And to present the clinical case of a patient affected by pleomorphic adenoma in the minor salivary gland.

MATERIAL AND METHODS

A case report will be presented and an integrative literature review will be carried out, from the electronic databases: *Medline* via

Pubmed, Scientific Electronic Library Online (Scielo) and Scientific and Technical Literature of Latin America and the Caribbean (LILACS). The following keywords were used: "Minor Salivary Gland Pleomorphic Adenoma"; "Pleomorphic Adenoma"; "Salivary Gland Tumor". Publications in Portuguese, Spanish and English were included. The following exclusion criteria were used: articles with restricted or private access, those whose titles or abstracts demonstrate that they are not useful for this research, and even those that presented limitations in the chosen topic.

LITERATURE REVIEW

SALIVARY GLANDS

The major salivary glands are made up of three glands, known as the submaxillary, sublingual, and parotid glands. They have the functions of secreting and producing saliva. They reach the oral cavity through *Stensen 's duct* (parotid gland) and *Wharton 's duct* (submaxillary glands) (MILORD, 2021).

Normally, it is produced and secreted from 1 to 1.5 liters of saliva in 24 hours, the submaxillaries are responsible for 71% of the seromucous type, the parotid for 25% of the serous type and the sublingual for 4% of the mucous type (MILORD, 2021). Through the minor glands, saliva is distributed throughout the oral mucosa. Inside these glands, numerous pathological processes can develop (SILVA, 2020).

Neoplasms in minor salivary glands are rare, but it is considered the second most common region to find possible lesions, studies show that the smaller the affected salivary gland, the greater the chance of malignancy in this region (PEREIRA, 2021).

INVOLVEMENT OF THE SALIVARY GLANDS

The region most affected by minor salivary gland tumors is the palate region, due to the greater accumulation of glands in this region. Another most affected region is the upper lips, followed by the jugal mucosa (NEVILLE, 2016).

Although tumors that reach the salivary glands are classified as the same group, they are clinically and morphologically different, they are differentiated through histopathological and differential diagnosis (SILVA, 2020). Tumors are made up of a combination of glandular epithelium and myoepithelial cells, which can form ducts and/or cystic structures, forming lesions (SILVA, 2020). The most common carcinoma in this region is mucoepidermoid, with adenoid cystic carcinoma and polymorphic adenocarcinoma also being classified in this class.

PLEOMORPHIC ADENOMA

Pleomorphic adenoma is considered a mixed tumor because it presents variable myoepithelial characteristics such as ductiform structures and a stroma of chondroid, hyaline, adipose, myxoid, bone tissue, containing a fibrous conjunctiva capsule. In different regions of the tumor, these variations can be seen, therefore pleomorphic- by various forms of presentation (BARROS, 2021).

Adenomas classified as pleomorphic are characterized by being a nodular, painless lesion that grows slowly, presents mobility and has well-defined limits. The superficial lobe in the preauricular and retromandibular region is the most affected region, both sides of the lobe are equally affected (PEREIRA, 2021). In cases of pleomorphic adenoma in minor salivary glands, the predominant region is the hard palate (SILVA, 2020).

The adenoma can affect people of any age group, including adults and young people, between 30 and 60 years old, has a predilection for females and presents a firm increase in volume. The lesion is round in shape and has a smooth surface, if trauma develops in

the lesion, an ulceration may occur in the region. When developed in the hard palate region, they are not considered mobile due to the strong adhesion to the tumor (NEVILLE, 2016).

DIAGNOSIS

The diagnosis, in addition to being clinical, can be performed by means of sialography, a method used to radiograph the salivary glands, ultrasound to confirm whether the lesion is cystic or solid, and computed tomography (CT) - actual size of the lesion, magnetic resonance - malignancy or benignity (BARROS, 2021).

In cases of pleomorphic adenoma located on the palate, radiographic exams will help little in the diagnosis due to overlapping bone structures, the exam that will stand out will be magnetic resonance imaging that offers a good image of soft structures (RODRIGUES, 2018). Lesions on the palate can affect the underlying bone, characterizing the well-defined transparent region in radiographic examinations, and may extend to the maxillary sinus (OLIVEIRA, 2016).

In this tumor, its diagnosis through histopathology, the slides are stained with hematoxylin and eosin and show the connective tissue and epithelial tissue (BARROS, 2021). The diagnosis can be discovered through fine needle aspiration (FNA) and excisional biopsy (SILVA, 2020).

The lesion, through histopathology, is usually encapsulated and well delimited, but the capsule can demonstrate infiltration, the demonstration of the imperfect capsule is common in tumors with involvement of the minor salivary gland, mainly in the external region of the palate, which is located below of the epithelial surface (NEVILLE, 2016).

There is wide variation among neoplasms due to the distribution of epithelial elements and mesenchyme components. The composition of the parenchyma includes glandular epithelium and myoepithelial cells. Histopathologically, we can observe ducts and cystic structures, keratin and mucus producing cells (NEVILLE, 2016).

BIOPSY

Biopsy refers to the collection of altered tissues in order to close the diagnosis during an anatomopathological analysis. Analysis is the main surest method for diagnosing some injuries. Excisional biopsy is recommended for small lesions smaller than 1cm in diameter, pedunculated or well-circumscribed lesions, such as papillomas, traumatic fibromas and others.

To perform the biopsy on intraoral structures, we need some instruments such as the carpule, anesthetics, needles, scalpel blades, scalpel handles, *Metzenbaum scissors,* Adson forceps, hemostatic forceps, field forceps, Minnesota retractor, needle holder forceps, suture thread, surgical sucker, gauze and a pot containing 10% formaldehyde. In some cases, a reamer, a spherical drill and a Lucas curette are required. The sample must be sent to the laboratory in the pot containing formaldehyde, without tissue lacerations (BRAZAO-SILVA, 2018).

DIFFERENTIAL DIAGNOSIS

Its differential diagnosis is based on lesions such as: mucocele, lipoma, fibroma, mucoepidermoid carcinoma, cystic adenomatoid carcinoma, acinar cell carcinoma and adenomacarcinoma (BARROS, 2021).

TREATMENT

Because it has a close relationship with the facial nerve, the removal of benign tumors is a case to be discussed in detail. In cases where the tumor reaches the parotid gland, the forms of treatment to be considered are: enucleation, superficial and total parotidectomy, it varies

according to the location of the tumor. In cases of involvement of minor salivary glands, the safest form of treatment and with a lower rate of recurrence is conservative enucleation of the lesion (PEREIRA, 2021).

Enucleation consists of removing the tumor in a way that does not remove glandular tissue. In cases where the surgeon opts for superficial parotidectomy, the tumor is removed with a portion of the salivary gland that is congregated to the tumor. In advanced cases, when the total is chosen, the entire gland will be removed, only the facial nerve will be preserved (RODRIGUES, 2018).

Cases of pleomorphic adenoma that recur are due to incomplete removal of the lesion or gland, in situations where it is already compromised (PEREIRA, 2021). When the capsule ruptures, it may increase the chances of recurrence. This way, the appropriate surgical technique becomes necessary (RODRIGUES, 2018).

COMPLICATIONS

Postoperative complications that may occur are: hematomas - due to poor hemostasis, unsatisfactory drainage, improper compression; infections - due to poor hygiene of the surgical wound; seroma or salivary fistula-outflow of saliva through the skin; *Frey* 's syndrome - sweating and flushing of the cheeks; and/or injury to the facial nerve- may cause paralysis or hypofunction of a specific branch (PEREIRA, 2021).

In the vast majority of cases, enucleation is the form of treatment chosen due to the lower risk of complications, as it is a shorter and simpler surgery (PEREIRA, 2021). In cases of involvement of the palate, the treatment consists of subperiosteal exeresis by displacing the lesion from its insertion in the palate and periosteum, together with the involved mucosa (RODRIGUES, 2018).

When not treated properly, the lesion may evolve, and become a malignant lesion, such as adenomatoid carcinoma, aggressive and malignant lesion, this evolution is rare in minor salivary gland tumors, it is more common neoplasms that affect the parotid (RODRIGUES, 2018).

Early diagnosis and correct management of structures in the proposed treatment are key factors for a positive prognosis, requiring a great contribution from the surgeon regarding the knowledge of anatomy and physiology of the glands involved (GAMA, 2018).

CASE REPORT

Patient MLA, female, black, 38 years old, born in Recife, was referred to the dental service for evaluation of intraoral swelling. In the anamnesis, he reported as the main complaint volume increase, slow growth and lasting 03 years. He had already looked for another dentist, who advised the mouthwash with Periogard and discharged the return. On extraoral physical examination, the patient presented a symmetrical face, mouth opening and cervicofacial ganglionic chains without alterations. In the intraoral physical examination, normal colored mucosa was observed, but with the presence of a soft consistency tumor, measuring approximately 2.3 cm in diameter, in the transition between the hard and soft palate.

The hypotheses of clinical diagnoses were mucocele and salivary gland neoplasms. With all preoperative exams normal, an incisional biopsy of the lesion was performed, which revealed it to be a pleomorphic adenoma.

The proposed treatment was removal of the lesion, with safety margins, under general anesthesia, due to the posterior location of the neoplasm. Despite this, local anesthetic infiltration with 2% lidocaine associated with 1:100,000 epinephrine was

also performed to control bleeding. The lesion was removed subperiosteally with a *Bard-Paker scalpel,* fitted with a number 15-c blade, associated with cauterization of the bloody region with an electric scalpel plus use of topical hemostatic, thus promoting better hemostasis, since, due to the size of the lesion, secondary intention healing would occur.

Nimesulide 100mg every 12/12 hours for 3 days, dipyrone 500mg every 6 hours for 3 days and Amoxicillin 500mg every 8 hours for 7 days were prescribed postoperatively. In addition to post-surgical guidance, such as cryotherapy, cold and pasty food for 3 days, guidance was given regarding cleaning the surgical wound with 0.12% Chlorhexidine Gluconate, after 24 hours for 7 days.

The patient recovered without any post-surgery intercurrence and is being followed up to the present day with no signs of lesion recurrence. Images of the procedure in figure 1.

Fig. A- Clinical appearance of the lesion. Fig. B- Surgical piece. Fig. C- Immediate postoperative period. Fig D.- Plate made to aid in the healing of the surgical wound

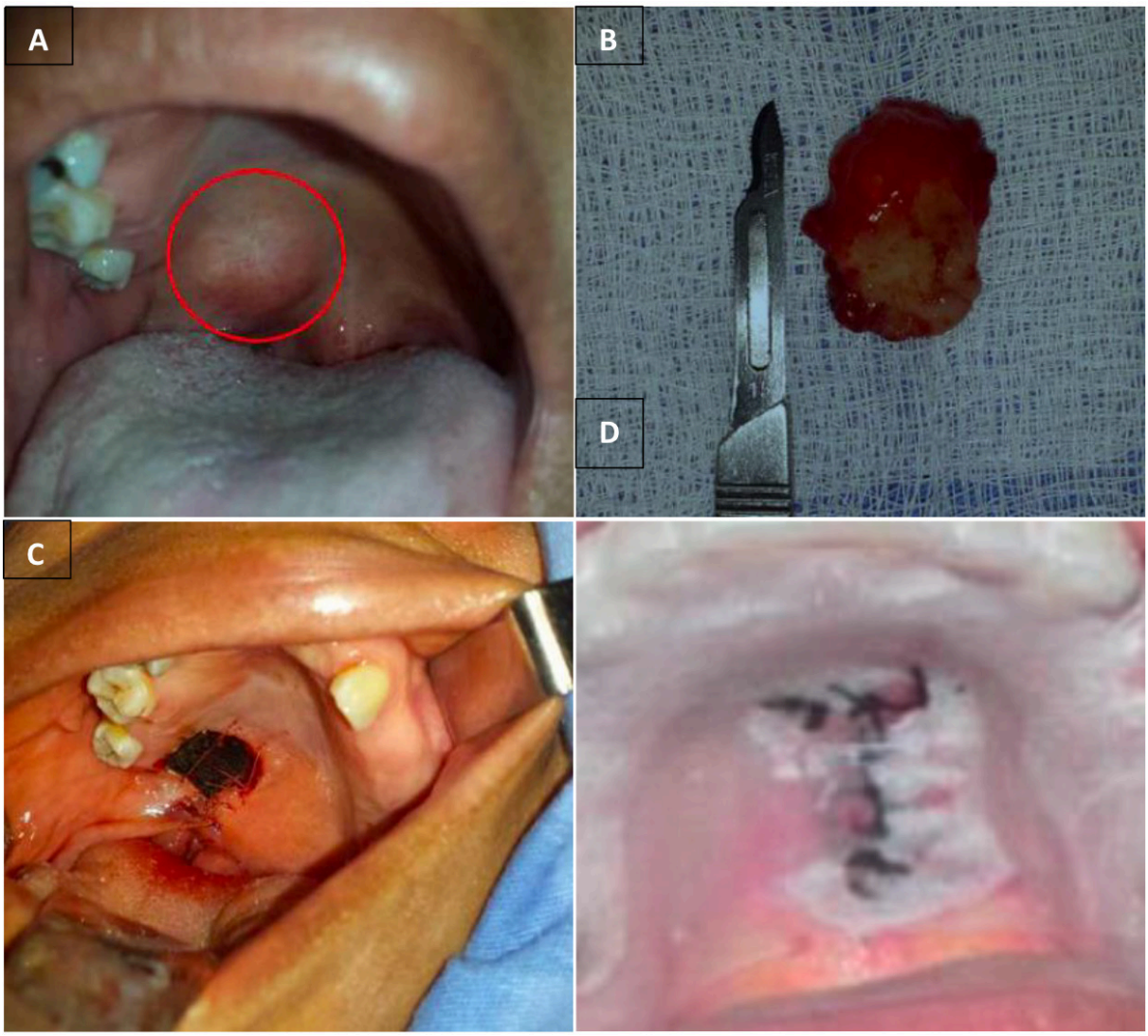

Figure 1. Preoperative and Postoperative Aspects.

Source: The author (2022).

DISCUSSION

Pleomorphic adenoma preferentially affects female patients (60%) between the fourth and sixth decades of life. The parotid gland is the most affected (RODRIGUES, 2018). Among the minor salivary glands, the palate region is most frequently affected. In the present case, the patient presents age, sex and anatomical location according to the literature.

Although minor salivary gland tumors are considered rare, the palate region is the most affected, involving head and neck tumors, however, there are few reports in the literature about these cases. According to Neville (2016), the most common region of involvement is the palate area, followed by the upper lips and buccal mucosa. According to Gama (2018), there is a predilection among women, with no defined age, however, a greater occurrence between the fourth and sixth decade of life is observed. Clinically, they prove to be firm, mobile to palpation, well delimited, without painful symptoms and of slow growth.

Due to Pleomorphic Adenoma presenting itself as a slow growing and painless lesion, its diagnosis often becomes late. In the present case, the patient took about 3 years to have the definitive diagnosis. Due to the variation in the clinical characteristic, which every pathology has, the growth of the lesion can also be developed quickly, present great extension and ulcers in the tumor region (OLIVEIRA, 2016). Therefore, a clinical and attentive look by the dentist is of great importance in the face of any injury. Favoring not only a faster diagnosis, but also the patient's prognosis.

The analysis of the pathology is extremely important so that there is a correct diagnosis, in order to carry out a good planning, approach and correct resection if necessary (HENNA, 2021).

Microscopically, the samples are stained with hematoxylin and eosin, and show epithelial and myoepithelial cells, surrounded by a fibrous tissue capsule, through histological characteristics, it is possible to diagnose the lesion (OLIVEIRA, 2016). Lesions that develop in the palate region are capable of reaching underlying bones and may expand to the maxillary sinus area. The diagnosis can also be given through radiological images, such as computed tomography and magnetic resonance (OLIVEIRA, 2016).

The differential diagnosis is through odontogenic cysts, abscesses located in the palate, fibroma, lipoma, among others. The imaging exams most used in management planning consist of computerized topography and magnetic resonance imaging to visualize the biological invasion of the lesion and the precise anatomical location. Being able to prevent intercurrences and change of plans in the planning of the surgical approach and removal of the lesion (COSTA, 2020).

Due to the slow growth of the lesion, there is a delay in seeking the dentist, which can delay the diagnosis and prognosis, as the lesion can evolve into a malignant lesion. The professional's lack of information and knowledge will also affect the patient's prognosis. Since, the reported patient, sought the dentist and he reported that he did not need to worry, and discharged the patient. The search for knowledge must be carried out constantly, the difficulty of making a diagnostic hypothesis may lead to greater problems for patients. Contributing to the evolution and malignancy of pathological lesions (GOMES, 2022).

The tumor in question has the capacity to transform into a malignant one, with about a 5% chance, the removal of the lesion, when located in the palate, must be performed below the periosteum covering the adjacent mucosa, involving a safety margin, in order to avoid recurrence (BUENO, 2018).

In the clinical case addressed, an incisional biopsy was performed to confirm the diagnostic hypothesis, followed by removal of the lesion with a safety margin, subjecting the patient to healing by second intention due to the location of the adenoma. The lesion must be removed with a safety margin so that there is no risk of remaining remnants of the lesion and consequently causing recurrence. Even if the chance of malignancy is considered low, due measures and correct conduct must be taken to avoid them (FLORES, 2018). In the case in question, due to good conduct, planning and effectiveness in the procedures performed on the patient, she is recovering well and without recurrence of the lesion.

FINAL CONSIDERATIONS

Minor salivary gland pleomorphic adenoma is considered rare and the diagnosis requires attention. The correct diagnosis, choice of histopathological analysis, good quality radiographic exams and a well-indicated surgical technique promote a better prognosis for the patient with a low probability of recurrence.

According to the literature, although the lesion is mostly benign, there are chances of becoming malignant, chances that may increase due to the negligence of professionals from diagnosis to treatment plan. It is noteworthy that the search for treatment quickly and preventively will contribute to early diagnosis, with higher rates of good prognosis.

Therefore, it is emphasized that the early diagnosis of pleomorphic adenoma is extremely important, in order to carry out adequate planning and avoid further consequences for the patient.

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