

Pleomorphic adenoma treatment in a patient with neurodegenerative disease: case report

Tratamento de adenoma pleomórfico em paciente com doença neurodegenerativa: relato de caso

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ABSTRACT

Pleomorphic adenoma (PA) is a benign tumor, responsible for more than 50% of the cases of salivary gland neoplasms, occurring both the major and minor salivary glands. In the oral cavity, the most frequent sites of pleomorphic adenoma are the palate and the lips, with greater preference for females and frequently affecting individuals in the fourth to fifth decade of life. The treatment of choice is based on total lesion excision with a good prognosis, which has a low potential for recurrence and malignant transformation. The objective of the study was reporting

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a treatment of pleomorphic adenoma localized in the hard palate of the 74-year-old male patient, F.A.S. with Parkinson's disease. The patient is in outpatient follow-up for over a year with no signs of recurrence. Computed Tomography is an indispensable tool for directing the correct diagnosis, which is only definitive after the histopathological examination. The most indicated treatment with good prognosis is complete surgical excision of the lesion and curettage.

Indexing terms: Oral diagnosis. Pleomorphic adenoma. Salivary Gland neoplasms.

RESUMO

O adenoma pleomórfico é um tumor benigno, responsável por mais de 50% dos casos de neoplasias de glândulas salivares, podendo ocorrer tanto nas salivares maiores quanto nas menores. Na cavidade oral, as localizações mais frequentes do adenoma pleomórfico são o palato e os lábios, tendo maior predileção pelo sexo feminino e, frequentemente, acomete indivíduos da quarta a quinta década de vida. O tratamento de escolha é baseado na excisão total da lesão com um bom prognóstico, o qual apresenta um baixo potencial de recidiva e de transformação maligna. O objetivo do estudo foi relatar o tratamento de adenoma pleomórfico localizado em palato duro do paciente F.A.S., 74 anos de idade, do sexo masculino portador de doença de Parkinson. O paciente está em acompanhamento ambulatorial por mais de 1 ano e sem sinais de recidiva. A Tomografia Computadorizada é uma ferramenta indispensável para o direcionamento do correto diagnóstico, o qual só é definitivo após resultado do exame histopatológico. O tratamento mais indicado e com bom prognóstico é a excisão cirúrgica completa da lesão e curetagem.

Termos de indexação: Diagnóstico bucal. Adenoma pleomorfo. Neoplasias das glândulas salivares.

INTRODUCTION

Salivary glands are anatomically classified into major and minor, of which the major salivary glands, especially the parotid glands, are the most affected by neoplastic processes [1].

Among the major benign tumors, Pleomorphic Adenoma (PA) accounts for more than 50% of salivary gland neoplasms. It is found in both major and minor glands (only 5-10% of cases). The parotid gland is the most commonly affected major gland. When it affects the minor salivary gland, AP can be located on the palate (mainly at the junction of the soft and hard palate), the lip, nasal cavity, pharynx, larynx, and trachea [2-5].

The highest prevalence of these tumors is related to female patients, in the fourth to fifth decade of life, but can occur in both sexes and at different times of life. A history of alcoholism, smoking, radiation, hormonal problems, and infections may increase susceptibility to the development of these oral neoplasms [6].

Parkinson's disease (PD) is a degenerative, progressive, incurable disease affecting mostly older people. As the main clinical features, patients present impaired motor systems, causing tremors and inflexibility and hindering the performance of common tasks such as oral hygiene. As prevalent oral symptoms, they present difficulty swallowing, complaints of xerostomia, burning mouth, problems with the use of dental prostheses, caries, and periodontal disease. In this context, the dental surgeon has an important role in minimizing the limitations presented by the patient, as well as diagnosing and treating possible oral lesions [7].

AP of the oral cavity (of the minor salivary glands) is most prevalent on the palate. The clinical features are a firm, unilateral, smooth-looking bulging of the covering mucosa, usually with slow growth.

Normally, the lesion is painless, with no infiltrating power into underlying structures, and is covered by the integral, normally colored oral mucosa [8,9].

Well-defined radiolucent areas, formed by compression resulting from the slow growth of the neoplasm, can be seen on complementary imaging exams (X-rays, CT scan, ultrasound, MRI). Histopathological examinations show encapsulated cells of well-defined fibrous connective tissue with epithelial, myoepithelial, and mesenchymal elements of complex morphology. There are also myxoid, hyaline, osteoid, plasmacytoid, ductiform, chondroid, solid, and even squamous metaplasia areas. For this reason, the tumor is called pleomorphic [3,9,10].

Despite the low risk of recurrence, the recommended treatment is total surgical excision of the PA. When complete removal of the benign tumor is performed, with no residue remaining from the capsule, it has a very favorable prognosis. Thus, the chances of recurrence and the potential for malignant transformation are greatly reduced [10-12].

The objective of this paper is to report a case of PA in an elderly patient with Parkinson's disease treated surgically under general anesthesia, addressing the diagnostic process and the surgical method.

CASE REPORT

F. A. S., a 74-year-old male patient, melanodermic, living in a rural area, with a medical history of Parkinson's disease, was referred to the Oral and Maxillofacial Surgery and Traumatology Department of the Hospital Universitário de Sergipe (HU/UFS) due to the presence of a nodular lesion on the hard palate.

The lesion was painless, with slow and progressive growth, noticeable for approximately one year. The intraoral clinical examination (figure 1) showed a nodular bulging on the palate, 3.5 cm in diameter, of firm and immobile consistency, with well-defined limits. It was located on the right posterolateral region of the hard palate, with extension to the soft palate, with an initial diagnostic hypothesis of Pleomorphic Adenoma (PA).



Figure 1. Clinical aspect of Pleomorphic Adenoma located on the palate.

Computed tomography (CT) imaging revealed a defined, unilocular, hypodense image affecting the alveolar bone in the regions corresponding to teeth 18 and 17, with bulging (figure 2) and disruption of the inferior cortical bone of the maxillary sinus, with disruption of the floor of the nasal fossa and resorption of the palatal cortical bone (figure 3).

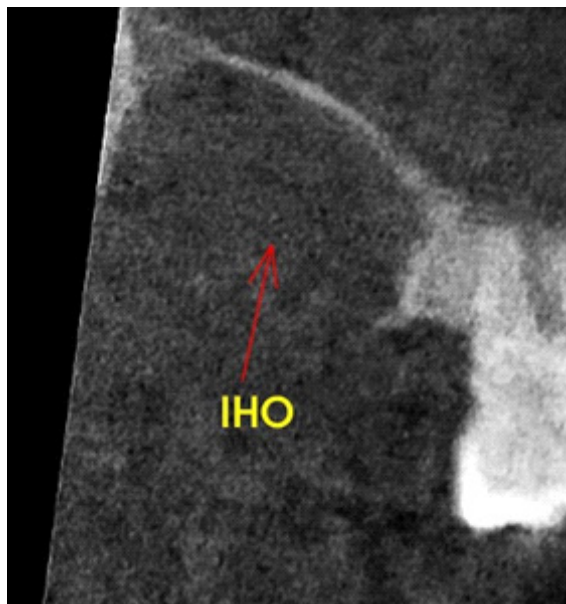


Figure 2. Cone beam Tomography (sagittal view). Presence of a defined, unilocular, hypodense image (IHO) affecting the alveolar bone in the regions corresponding to teeth 18 and 17 with bone bulging.



Figure 3. Cone beam Tomography (coronal view). Disruption of the inferior cortical of the maxillary sinus, the floor of the nasal fossa and resorption of the palatal cortical bone (arrow).

Total surgical excision of the lesion was performed under general anesthesia in the surgical center of the Hospital Universitário de Sergipe, and the specimen was sent for histopathological analysis (figure 4). Macroscopically, the anatomopathological result reported a nodular formation of capsular tissue, yellowish, unctuous, shiny, measuring 3.5 x 2.5 x 2.0 cm (figure 5). Histological analysis showed a fragment of benign salivary gland neoplasm characterized by ductiform arrangements of epithelial cells and variety of stromal component (myxomatoid, chondroid and hyalinized). The microscopic picture was compatible with Pleomorphic Adenoma and the morphological analysis identified the complete removal of the lesion.

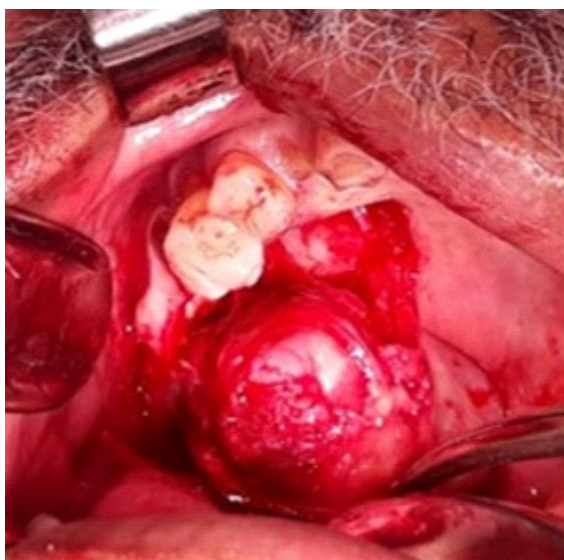


Figure 4. Transoperative appearance during surgical excision.



Figure 5. Specimen sent for anatomopathological evaluation.

Patient follow-up was carried out at the outpatient clinic of the Unit of Oral Diagnosis and Dentistry for Special Patients (UDOPE) of the Hospital Universitário de Sergipe (HU/UFS). After a year of treatment, the patient is in good general condition, presenting the surgical site region with a normal healing pattern of the palatal mucosa and no signs of recurrence (figure 6).



Figure 6. Postoperative period after 1 year of clinical follow-up. Complete healing of the mucosa covering the palate.

DISCUSSION

The etiology of PA still remains unknown, but the incidence of this neoplasm has increased over the past 20 years. Studies show a possible association with radiation exposure, with radiation oncology treatment of the head and neck region also being a risk factor. Another important association suggested is the presence of the oncogenic simian virus 40 (SV 40) which has been related to the onset or progression of AP [1,6,10]. The patient in the present clinical case had no history of radiotherapy treatment, but exposure to natural radiation because he lived in a rural area and had a life of working in the fields. The health problems he presented, besides Parkinson's disease, were heart disease and hypertension, but well controlled.

Of all the salivary gland tumors, PA represents an incidence between 45 and 75% of cases, being the most common benign neoplasm of glandular origin. It is observed in individuals of all ages, but is more prevalent after the third decade of life, and affects a higher proportion (2:1) of women [1,12]. The parotid gland has been the most frequent site of AP occurrence [3,4]. When it affects smaller glands, 5 to 10% of cases, it occurs mainly in the soft and hard palate due to the higher concentration of these gland types [3,4,6,12]. The patient in the present clinical case was 74 years old, i.e. in the most affected age group according to the studies mentioned, the third decade of life on. In addition, he had the lesion in a minor salivary gland, in the region of the hard palate with extension to the soft palate. Regarding the prevalence in sex, as a male patient, he was out of the predominance reported by most studies. However, Patigaroo et al. [13] reported that PA located in palate has predominance in male over female gender, however, with the most common age range between 16 and 30 years old.

The lesion of the presented case was painless, with slow and progressive growth, noticeable for approximately one year. On clinical examination, it was nodular in shape, with 3.5 cm in its largest diameter,

firm and of immobile consistency, with well-defined limits, in agreement with most studies [1,6,13]. Some cases of PA in the palate region may present ulceration, pain and bleeding probably due to repeated trauma when chewing [13]. In addition, in minor salivary gland tumors, other symptoms can be reported, such as hoarseness, dysphagia, dyspnea, difficulty chewing, and epistaxis. Rapid enlargement of this tumor nodule is uncommon and should raise a suspicion of malignant change [1,6,14]. In the clinical case presented, although the patient reported no pain or bleeding, the lesion was ulcerated, probably due to masticatory trauma. Difficulty in swallowing due to the large extension and more posterior location on the palate was also reported. However, slower growth and benign characteristics have been observed.

Confirmation of the tumor is done by Computed Tomography (CT) and Magnetic Resonance Imaging.

The clinical examination complemented by imaging exams, such as CT and magnetic resonance imaging, provide important information about location, size, and extent of the tumors [1,3,6]. Usually, the image appears as a hypodense globular mass with smooth or lobulated margins with few calcification points. CT was the imaging exam of choice in this clinical case because of the concern to evaluate the possibility of compromising the bony structure of the palate. Thus, a hypodense, unilocular image with defined margins was observed, affecting the regions between teeth 17 and 18. Through the tomography, it was also possible to diagnose the bulging with discrete rupture of the inferior cortical of the maxillary sinus, besides the slight involvement of the floor of the nasal fossa. CT images showed features that can be observed in the study by Patigaroo et al. [13]. The authors reported that in cases of PA on the palate, involvement of the bony wall is uncommon, except in cases with a prolonged time of evolution. This fact was observed in the present clinical case, which had an evolution of more than a year.

Currently, local wide excision of the lesion, involving its capsule, and curettage of the involved bone is the most accepted form of treatment [1,6,9,10,12,13,15]. Surgical manipulation with incomplete removal of the tumor has the potential for hematogenous spread and is related to recurrence [1,10]. Complete excision, when done correctly, has a cure rate of around 95% [6,9]. In this sense, the case reported in the present study had total excision as the surgical option. The patient has been followed for over a year without recurrence.

Microscopically, the PA is widely variable, hence the name pleomorphic, making histopathological diagnosis challenging. It is composed of mixed proliferation of polygonal epithelial cells and fusiform myoepithelial cells in a variable stromal matrix, and can be of mucoid, myxoid, cartilaginous, or hyaline origin [1,6,9,14]. In the present study, the histopathological report showed a nodular formation of capsular, yellowish, unctuous, shiny tissue. In addition, ductiform arrangements of epithelial cells and stromal component (myxomatoid, chondroid and hyalinized) stood out. The microscopic picture was compatible with AP and the morphological analysis identified the complete removal of the lesion.

When dissemination of PA occurs, the benign characteristic is rarely preserved, and malignant transformation is the main problem to be investigated [10]. However, PAs have small malignant potential (on average 6%) and is proportional to the length of time the tumor has been around. Other risk factors for malignancy include advanced age, radiotherapy treatment, and recurrent tumors [1,3,6,12,16]. Although recurrences are uncommon after adequate surgical treatment, the case must be followed up on a long-term basis [13]. From the beginning, the patient reported had benign characteristics for the PA on the palate, and no signs of malignancy were observed on histopathology, nor was there any dissemination or recurrence of the lesion in the post-operative follow-up.

No studies have been found in the literature that correlate Parkinson's disease with Pleomorphic Adenoma or any other tumor of the oral cavity. Typically, the oral clinical features of this neurological condition include: difficulty swallowing, complaints of xerostomia, burning mouth, problems with dentures, tooth decay, and periodontal disease due to motor problems. In this context, the dental surgeon has an important role in minimizing the limitations presented by the patient, as well as diagnosing and treating possible oral lesions, such as the pleomorphic adenoma described in this study.

CONCLUSION

AP on the palate is not a common lesion, usually slow growing and painless. CT is an indispensable tool to guide the correct diagnosis and evaluation of possible cortical bone disruptions. Definitive diagnosis occurs by histopathological examination. The most indicated treatment with good prognosis is complete surgical excision of the lesion and curettage.

Collaborators

ILS Maia, conceptualization (equal), data curation (equal), writing- original draft (equal), writing- review & editing (equal). PPS Santos, review & editing (equal). AC Marqueti, writing- review & editing (equal). PHL Freitas, surgical execution and manuscript review (equal). RLB Oliveira and AB Cardoso, conceptualization (equal), project administration (equal), writing- original draft (equal), writing- review & editing (equal).

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