

Unusual treatment of pleomorphic adenoma of soft palate in adult

Tratamiento inusual del adenoma pleomórfico del paladar blando en adulto

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ABSTRACT: Pleomorphic adenoma is the most common benign neoplasia of the salivary glands and affects mostly the parotid gland, less frequently the minor salivary glands. Minor salivary gland tumors have a higher risk of malignancy compared to tumors of the major salivary glands, so appropriate diagnostic evaluation should be prompt. In this case report, we present a case of an extensive pleomorphic adenoma of soft palate in an adult patient. After preoperative investigation using Fine Needle Aspiration (FNA) and imaging tests, the patient was successfully treated by surgical resection under general anesthesia. There was no recurrence seen after a follow-up period of 1 year.

KEY WORDS: Pleomorphic adenoma, minor salivary gland tumor, palate, surgical treatment.

INTRODUCTION

Pleomorphic adenoma is the most common benign neoplasia of the salivary glands, with 70% of cases occurring in the parotid gland of slow and painless growth (Antunes & Antunes, 2005; Andreasen *et al.*, 2018; Hmidi *et al.*, 2015). Approximately just 10% of pleomorphic adenoma occur in the minor salivary glands (Forde *et al.*, 2018; Passi *et al.*, 2017). However, when occur in the minor salivary gland the tumors have a higher risk of malignancy (Forde *et al.*, 2018).

The great extent of this intraoral tumor is not common and when it occurs there is preference for the palate. Other intraoral sites include lips, buccal mucosa, tongue, floor of mouth, tonsil, retro-molar area, pharynx and nasal cavity (Passi *et al.*, 2017). Despite there is no certainty about the origin or what causes the disease to the present moment (Antunes & Antunes, 2005), some genetic hypothesis will be discuss. In this paper we will present a case report of a treatment for an unusual adenoma located at the soft palate in an adult patient.

CASE REPORT

Male, 25 years old, attended at the Oral and Maxillofacial Surgery Department of Angelina Caron Hospital, Curitiba - PR, Brazil. The patient had a nodular lesion, single, asymptomatic, firm consistency, with a non-bleeding evolution period of approximately 18 months, located in the left hemisphere of the soft palate next to the hard palate in the molar area (Figure 1). Lesion was well delimited and has approximately 45 mm in diameter without ulceration or surrounding inflammation. No difficult to speech or swallowing were presented by the patient.

As initial management, magnetic resonance and videolaryngoscopy examinations were requested to define possible invasion of hard tissue injury and invasion of air space (Figure 2). The result showed very well delimited lesion, with preservation of the adjacent structures without invasion to the nasopharyngeal space.

As a preoperative exploration, surgical procedures were performed, doing Fine needle aspira-

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Fig. 1. Inicial image of the tumor on the palate.

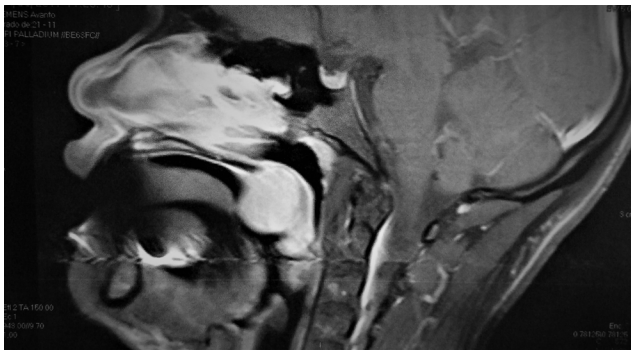


Fig. 2. Magnetic resonance imaging showing the lesion in the retro pharyngeal area, impairing the superior airways.

tion (FNA) to exclude some degree of malignancy and subsequent surgical resection in operating room under general anesthesia was proposed. Fine needle aspiration (FNA) is an excellent diagnostic method for the analysis of malignancy or not of tumors linked to salivary glands, but does not present specificity in the analysis and sampling of the cells present in the lesion [3]. The result of FNA showed no malignancy in this case.

Prior to the surgical procedure, a Hawley type restraint plate was performed to obliterated the local area to avoid surgical wound injury (Figure 3). Patient underwent surgical procedure under general anesthesia to remove total injury. After orotracheal

intubation and local anesthesia with vasoconstrictor, the incision at the anterior border of the lesion was performed, followed by delicate detachment and divulsion by planes with the purpose of maintaining the integrity of the pathology and favoring histopathological examination, as well as sufficient tissue for surgical closure by primary intension. The total removal of the lesion was made to confirm the diagnostic hypothesis. After, the suture with vycril 4-0 finalized the procedure (Figure 4, 5, 6, 7, 8). The laboratory examination confirmed after 2 weeks the diagnosis of unusual pleomorphic adenoma of soft palate.

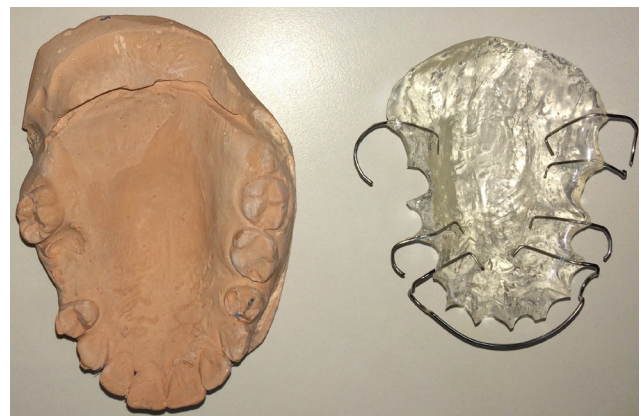


Fig. 3. The Hawley type restraint plate.

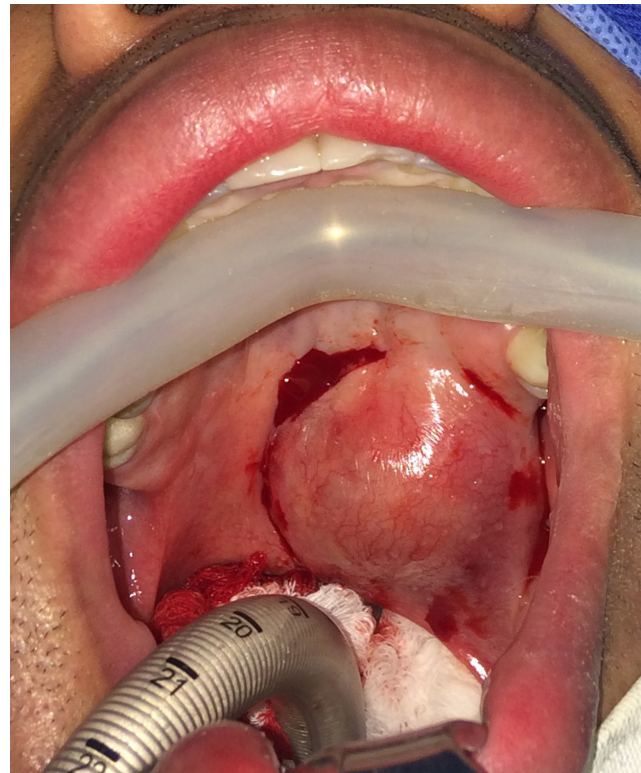


Fig. 4. The initial incision to resect the tumor.

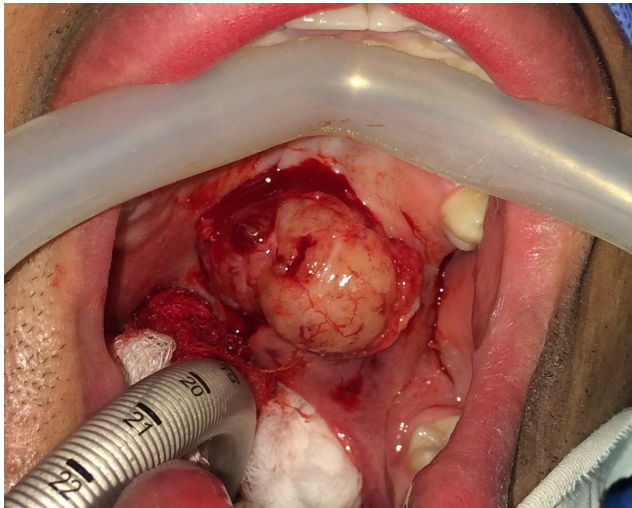


Fig. 5. Tumor exposure after dissection.

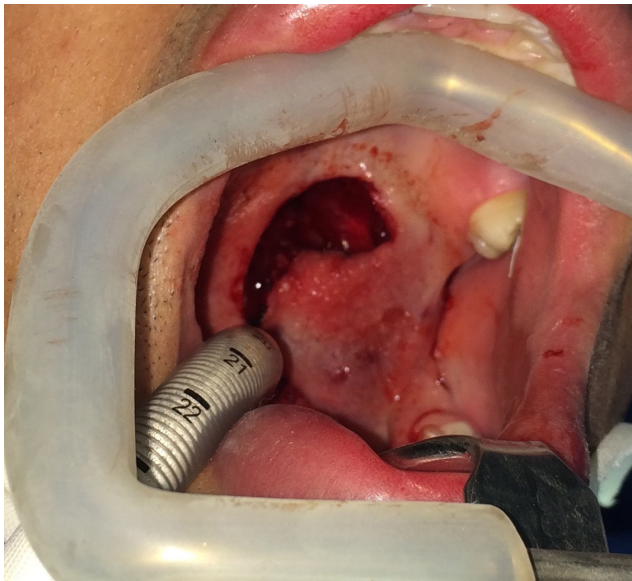


Fig. 6. Surgical site after total tumor resection.

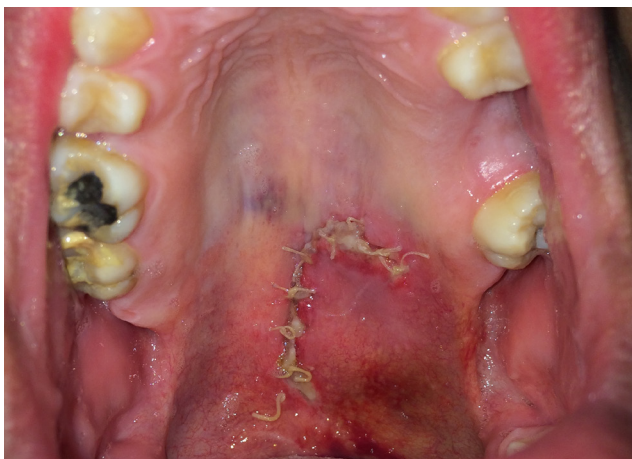


Fig. 7. Final suture in the first step.

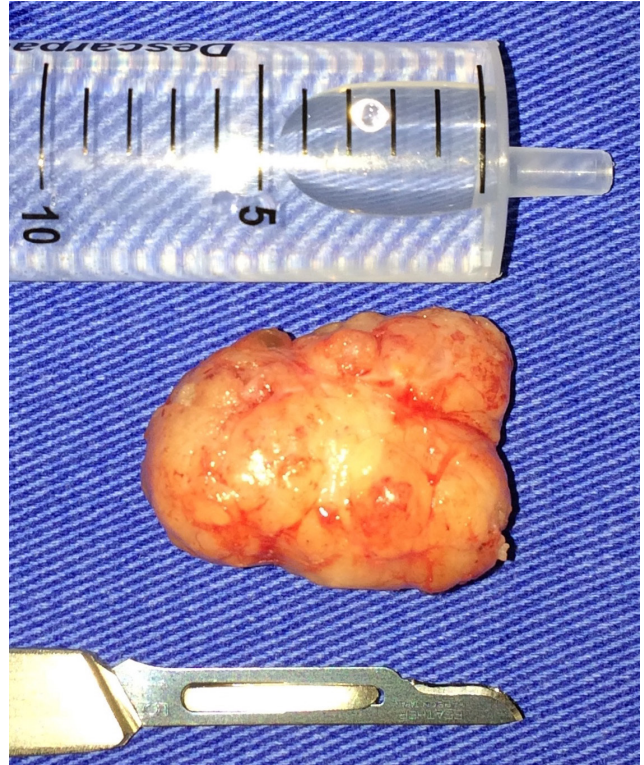


Fig. 8. Surgical tumor removed.

The patient remained under observations for 24 hours. At this time of recovering he presents a time of evolution of 1 year and continues to be followed up every 6 months for analysis of possible recurrence of the lesion (Figure 9).



Fig. 9. After 1 year of surgery, without signs of relapse.

DISCUSSION

According to World Health Organization (WHO), parotid gland neoplasms present about 30 subtypes (Silvers & Som, 1998), with pleomorphic adenoma being the most common of these

variations, representing around 60 to 70%, higher incidence from the 4th to the 6th decade of life (Seifert & Sobin, 1991, Passi *et al.*, 2017) and slight female preponderance (Forde *et al.*, 2018). It is the most common salivary gland neoplasm in children, representing 66-90% of all salivary gland tumor (Rahnama *et al.*, 2013).

Adenocarcinoma Polymorph, adenoid cystic carcinoma and Pleomorphic adenoma are the lesions that constitute the majority of tumors involving a palatal nodular mass. Considering different degrees of aggression, histological analysis is extremely important for surgical planning, approach and resection. Histologically, the adenoma is composed of ductal and epithelial cells based on a chondromyxoid stroma presenting malignant mutation for carcinoma (Andreasen *et al.*, 2018). Also, the term of pleomorphic adenoma is used to indicate the presence of both epithelial and mesenchymal tissues (Passi *et al.*, 2017). Research has shown epithelial origin of the mixed tumor, as well as clonal chromosome abnormalities with aberrations involving 8q12 and 12q15. The tumor often displays characteristic chromosomal translocations between chromosomes #3 and #8. This causes the PLAG gene to be juxtaposed to the gene for b-catenin. Thus, this activates the catenin pathway and leads to inappropriate cell division (Passi *et al.*, 2017; Rahnama *et al.*, 2013).

The differential diagnosis includes palatal abscess, odontogenic and non-odontogenic cysts, soft tissue tumors such as fibroma, lipoma, neurofibroma, neurilemmoma and lymphoma as well as others salivary gland tumors (Rahnama *et al.*, 2013).

Mark A Cohen reported in their study 144 cases of pleomorphic adenoma being 85% of the tumors located in the hard-palatal region, having a dimension of 10mm to 70mm in diameter (Cohen, 1986). These lesions are clinically isolated, solitary, mobile, except when it occurs on the palate, presenting slow and asymptomatic growth with variable dimensions, similar to the adjacent mucosa and firm to palpation (Silvers & Som, 1998; Forde *et al.*, 2018; Passi *et al.*, 2017). Ma Jaber studied 75 cases of the tumor in different parts of the oral cavity, being 26 in the palate, these presented malignancy in 42.3% (Jaber, 2006). Various large sizes of this tumor of soft palate have been reported in literature measuring since 1.9 x 2.0 cm until 4.5 x 3 cm (Passi *et al.*, 2017). Our case measured

about 4.5 x 5 cm and had been located at the soft palate, in a 25 years old male patient; which both parameters make this case unique.

The magnetic resonance imaging and computed tomography in the analysis of pleomorphic adenoma is important due to the observation of the biological invasiveness of the lesion, high contrast between the tissues and precise anatomical location (Andreasen, 2018; Rotta *et al.*, 2003; Forde *et al.*, 2018; Passi *et al.*, 2017; Rahnama *et al.*, 2013). Also, the FNA evaluation present a reasonably at excluding malignant disease in order to allow more accurate surgical planning (Pantanowitz *et al.*, 2018; Forde *et al.*, 2018). In the present case, we observed with the imaging exams that the lesion did not present adjacent anatomical structures that could lead to complications during the surgical resection procedure.

In the case presented, the complete resection of the lesion was performed after FNA, resulting in benignity, currently with 1 year of follow-up without relapses. According to Chidzonga *et al.* and Ford CT *et al.* conservative surgical excision is the ideal treatment for the pleomorphic adenoma of minor salivary glands having a low rate of relapse if removed correctly (Chidzonga *et al.*, 1993; Forde *et al.*, 2018). After incomplete surgeries, tumors derived from salivary glands tend to recur. Therefore, the best indication as primary treatment for these lesions is complete surgical excision with clinical and radiographic follow-up [Cohen, 1986; Jaber, 2006; Alves *et al.*, 2018; Hmidi *et al.*, 2015; Rahnama *et al.*, 2013]. Inadequate surgical procedure was reported to be the main cause of failure (Rahnama *et al.*, 2013). Relapses of lesions with longer evolution time increase the possibility of tumor malignancy (Ethunandan *et al.*, 2006).

CONCLUSION

To conclude, the pleomorphic adenoma of minor salivary glands is relatively rare and diagnosis requires attention. The correct early diagnosis for histopathological analysis, radiographic examination and correct surgical technique provide very good prognosis of treatment with low relapse rate. Recurrence after years as well as malignant transformation should be a concern and long-term follow-up is necessary.

Ethical responsibilities

Data confidentiality: The authors declare they follow the protocols of their work centers about the publication of patient's data.

Privacy rights and informed consent: The patient signed the informed consent and authorized the images publication.

Conflict of interest: The authors declare no conflict of interest.

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RESUMEN: El adenoma pleomórfico es la neoplasia benigna más común de las glándulas salivales y afecta principalmente la glándula parótida, con menos frecuencia en las glándulas salivales menores. Los tumores de las glándulas salivales menores tienen un mayor riesgo de malignidad en comparación con los tumores de las glándulas salivales mayores, por lo que la evaluación diagnóstica apropiada debe ser rápida. En este reporte de caso, presentamos un caso de un extenso adenoma pleomórfico de paladar blando en un paciente adulto. Después de la investigación preoperatoria utilizando aspiración con aguja fina y pruebas de imagen, el paciente fue tratado con éxito con la resección quirúrgica bajo anestesia general. No se observó recurrencia después de un período de seguimiento de 1 año.

PALABRAS CLAVES: Adenoma pleomórfico, tumor de glándulas salivales menores, paladar, tratamiento quirúrgico.

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