

# Palatal Pleomorphic Adenoma in a Pediatric Patient. Case Report

Adenoma Pleomorfo Palatino en un Paciente Pediátrico. Reporte de Caso

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**ABSTRACT:** Pleomorphic adenoma is the most common benign neoplasm of salivary glands. Their common location is in parotid gland, however, a lower percentage of these tumors might occur in minor glands. The epidemiology of this tumor shows that adults are the most affected, with rare occurrence in children or adolescents. We present the case report of pleomorphic adenoma located on the palate of a 10 year old. Excisional biopsy of the lesion followed by histopathologic examination of the biopsy specimen revealed ductal structures surrounded by plasmacytoid mioepithelial cells within a myxoid stroma, the final diagnosis corresponded to Pleomorphic Adenoma. Early detection and excision of this lesion in children are important to minimize potential recurrences or malignant transformation.

**KEY WORDS:** Pleomorphic Adenoma; Minor Salivary Glands; Salivary Gland Tumors.

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## INTRODUCTION

Pleomorphic adenoma (PA), also known as benign mixed tumor of salivary gland, is the most common benign tumor of salivary glands reaching up to 60 % of all salivary neoplasms (Ledesma-Montes *et al.*, 2002; Barnes *et al.*, 2005). The usual location is parotid gland, however a small percentage arises in minor salivary glands (Pusztaszeri *et al.*, 2009; Erdem *et al.*, 2011).

Clinical examination is key to identifying the PA, some of the characteristics of this tumor are: an ovoid shape, usually smooth surface, grayish-white colour and semi-hard consistency, although it can be of hard consistency because of the presence of chondroid or osseous tissue (Ledesma-Montes *et al.*). Among the complementary techniques to diagnose PA are the following: Magnetic Resonance Imaging (MRI), Computed Tomography (CT) alone or combined with sialography or puncture and Fine Needle Aspiration (Pons Vicente *et al.*, 2008). The use of these techniques could guide towards a definitive diagnosis, but no one replaces the biopsy as the gold standard for the diagnosis of this neoplasia.

Histopathological elements of the PA include: capsule, epithelial and myoepithelial cells, and mesenchymal or stromal elements (Barnes *et al.*). The etiology of PA's is unknown, however some authors mention the myoepithelial cell as responsible for the development of this pathology (Ledesma-Montes *et al.*). Seifert *et al.* (1976) mentioned that PA's can be divided according to the proportion of cellular and stromal elements within the tumor, and therefore be classified as myxoid, classic or cellular.

The aim of this study was to report a palatal pleomorphic adenoma in a 10-year-old child.

## PATIENT INFORMATION

A 10-year-old patient comes to the clinic of the Faculty of Dentistry, Mesoamerican University for pediatric dental treatment. Within medical history he does not refer to anything of importance. The intraoral examination showed a ovoid nodule, located in right posterior third of the hard palate, sessile base, soft

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consistency, a little paler than the adjacent mucosa, smooth surface, asymptomatic and unknown evolution (Fig. 1), no bone involvement was evident in the radiographic study. Under this description presumptive diagnosis of Pleomorphic Adenoma was made. Core incisional biopsy was made and the preliminary diagnosis was Pleomorphic Adenoma. Excisional biopsy of the lesion to confirm the diagnosis was indicated.

The patient was scheduled for surgical excision of the lesion. Under the effect of general anesthesia and nasotracheal intubation, the infiltration of 2 % lidocaine plus epinephrine 1:100,000 was performed around the lesion to acquire selective hemostasis. Next it was excised down to periosteum leaving a safety margin of 2 mm, complete resection of the lesion was achieved, obtaining a surgical specimen of 12 mm in

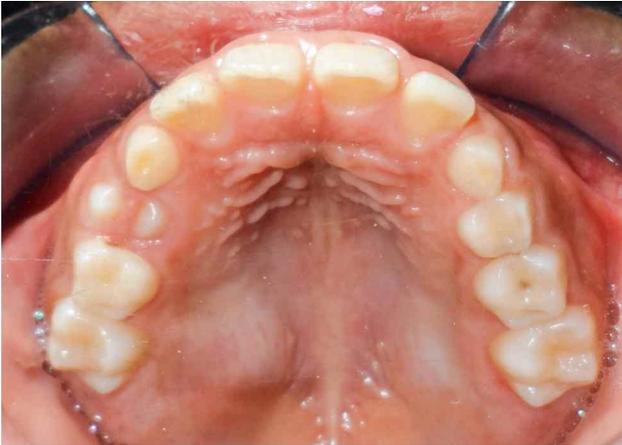


Fig. 1. Initial aspect of the lesion located in right posterior third of the hard palate.

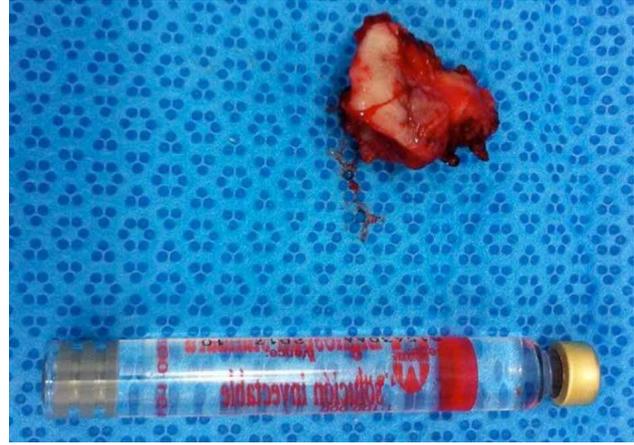


Fig. 2. Excised specimen.

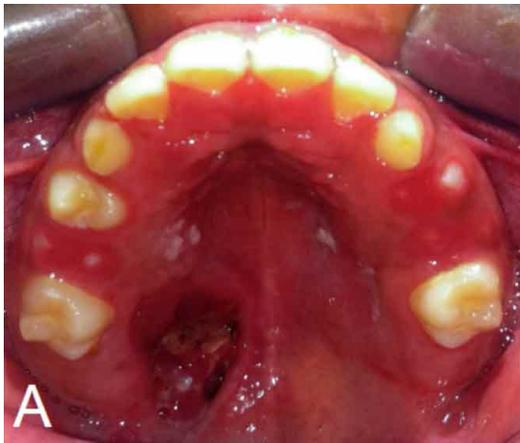


Fig. 3 (A.15 days postoperative reassessment, B. 30 days postoperative reassessment, C. 60 days postoperative reassessment).

diameter (Fig. 2). The surgical wound was bleeding so cauterization and hemostasis with hydrogen peroxide and pressure was performed, the wound was covered with surgical cement using an acrylic splint. Procedure was realized successfully. Patient with open mouth and is extubated without complications. The patient was reassessed at 15, 30 and 60 days after the procedure and did not show any post-operative complication (Figs. 3a, 3b, 3c). Histopathological results of the specimen sent presented the following findings: presence of a mixture of glandular epithelium and abundant islands with an aspect of plasmacytoid myoepithelial cells with embedded ductal formations in a myxomatous stroma, formation of fibrous capsule is not appreciated, but normal looking connective tissue surrounding the neoplastic tissue is evident (Fig. 4). It was determined as histopathological diagnosis Pleomorphic Adenoma.

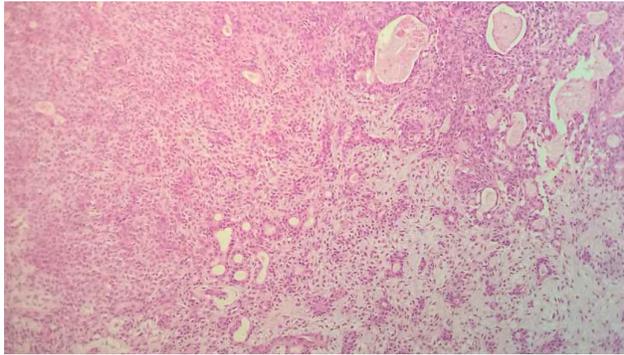


Fig. 4. Glandular elements embedded in mioepithelial cells surrounded by a myxomatous stroma (H&E 100x).

## DISCUSSION

Neoplasms of minor salivary glands in children are rare. It is mentioned that only 5 % of these have been reported in patients younger than 20 years (Moubayed *et al.*, 2010). The diagnosis of palatal PA is a real challenge because it can be confused with other entities such as odontogenic cysts or tumours, a palatal abscess, lymphoma, lipoma, fibroma, neurofibroma, neurilemoma (schwannoma), mucoepidermoid carcinoma or even cystic adenoid carcinoma (Hmidi *et al.*, 2015). The epidemiology of this neoplasm indicates that it is more common in female patients with a ratio of 2:1 in the fourth and fifth decade of life, in our case the patient was 10 years old which deserves a special mention since this neoplasia usually affects adults (Barnes *et al.*).

The origin of this neoplasm is still debated, some authors ascribe the origin of the PA to the reserve cell

of the intercalated duct, even so, it is possible that the myoepithelial cell also plays an important role in the development of this neoplasia (Ledesma-Montes *et al.*). Recent research shows that chromosomal translocations between chromosomes 3 and 8 may be responsible for tumor formation by the juxtaposition of PLAG-1 and b-catenin gene, resulting in the activation of catenin pathway causing cell proliferation (Rahnama *et al.*, 2013).

Histologically the PA can be classified into different types: myxoid, classic or cellular, depending on the amount of stroma and the proportion of cellular components containing. Myxoid type as its name suggests, is composed mostly of myxomatous stroma. The classic is a mixture of ductal structures and myoepithelial cell, and a myxomatous or hyaline stroma, The cellular type has a large number of cellular elements with little stroma (Wu *et al.*, 2016). Our case was cataloged as a classic PA due to the characteristics presented histologically.

Immunohistochemical profiles have been assessed for the diagnosis of PA's showing positive results for cytokeratin types 3, 6, 10, 11, 13, 16, S-100, Vimentin,  $\alpha$ -Smooth Muscle Actin, GFAP, Calponin, CD10, p63 and HHF-35 (Barnes *et al.*).

The patient was treated with intraoral conservative surgical resection with safety margins of the lesion and healing by second intention which is mentioned as the most ideal treatment (Barnes *et al.*; Alkan & Inal, 2008). It has been reported that radiotherapy as an adjuvant improves the outcome in PA's that have been resected with inadequate margins (Mendenhall *et al.*, 2008). Oroantral communication, recurrence and malignant transformation due to incomplete excision of the lesion are some of the possible complications mentioned in the treatment of palatal PA's (Shaaban *et al.*, 2001). None of these complications were observed in our case.

## CONCLUSION

Pleomorphic adenoma should be considered as a differential diagnosis when a mass appears in palatal portions of children. Biopsy and histopathological study are the gold standard to diagnose this pathology. Conservative surgical resection should be the treatment of choice to avoid recurrence or malignant transformation.

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**RESUMEN:** El adenoma pleomorfo es la neoplasia benigna más común de las glándulas salivales. Su localización común está en glándula parótida, sin embargo, un bajo porcentaje de estos tumores puede ocurrir en glándulas menores. La epidemiología de este tumor muestra que los adultos son los más afectados, con rara ocurrencia en niños o adolescentes. Presentamos el caso de un adenoma pleomorfo localizado en el paladar de un niño de 10 años. La biopsia excisional de la lesión seguida de examen histopatológico de la muestra de biopsia reveló estructuras ductales rodeadas por células mioepiteliales plasmocitoides dentro de un estroma mixoide, siendo el diagnóstico final adenoma pleomorfo. La detección temprana y la excisión de esta lesión en los niños es importante para minimizar las recidivas potenciales o la transformación maligna.

**PALABRAS CLAVE:** adenoma pleomorfo, glándulas salivales menores, tumores de glándulas salivales.

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