

# Pleomorphic Adenoma in a Young Female: Case Report

## Adenoma Pleomórfico en una Mujer Joven: Reporte de Caso

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**ABSTRACT:** Pleomorphic adenoma (PA) is the most common benign salivary gland neoplasm and it is frequently diagnosed in the third and fourth decade with predilection for women. PA is the most common benign lesion of minor salivary glands in children and adolescents, being the palate one of the most frequently affected site of minor salivary glands. Herein, we present a case report of a PA of the hard palate diagnosed in a 15-year-old female and a review of the English-literature of the reported cases of PA in children and adolescents in the hard palate.

**KEY WORDS:** mouth, hemangioma, vascular malformationboca, hemangioma, malformación vascular.

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### INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign salivary gland neoplasm and it accounts for 60% of these. It is frequently diagnosed in the third and fourth decade and has a predilection for women. In minor salivary glands, it generally occurs in the hard palate and in the upper lip. PA is the most common benign salivary gland tumor in children and mucoepidermoid carcinoma is the most frequent malignancy. Fewer than 5% of all primary salivary gland neoplasms occur in children, in the first decade of life, usually the first 2 years of life, with a preponderance of benign neoplasms (Seifert *et al.*, 1986; Callender *et al.*, 1992; Luna *et al.*, 1991; Jorge *et al.*, 2002; Mehta & Willging, 2006).

PA typically occurs as a firm, painless, and slow growing mass. In minor salivary glands, the palate is the most frequent location, and appears as a lobulated mass, usually covered by normal overlying mucosa. Histologically, it is characterized by the presence of epithelial and mesenchymal elements, giving rise to ductal structures, intermingled with a stroma with myxoid, hyaline, cartilaginous, and osseous change. This tumor is usually encapsulated, but extensions into adjacent tissues may be observed (Seifert *et al.*; Callender *et al.*; Luna *et al.*; Jorge *et al.*; Mehta & Willging).

Herein, we present a case report of a PA of the hard palate diagnosed in a 15-years-old female and a review of the English-literature of the reported cases of PA in children and adolescents with hard palate.

### CASE REPORT

A 15 year-old female patient was referred to the Oral Health Diagnostic Service with chief complaint of painless swelling for a period of six months, in the right posterior hard palate close to the first molar. Imaging exams did not show underlying bone involvement. The medical history was not contributory.

Clinical examination revealed a single swelling, enlarging the hard palate to the midline. The lesion is fluctuated slightly over the bone and showed a smooth surface with a color similar to the oral mucosa with telangiectasia-like appearance at the center (Fig. 1).

An incisional biopsy was performed under local anesthesia, without post-surgery complications. Macroscopically, was observed a pyramidal shaped, firm and smooth brownish-white specimen of 10.5 x 7

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x 6 mm of size. The histopathological examination noted a solid lesion covered by a orthokeratinized pluriestratified epithelium, surrounded by a fibrous capsule (Fig. 2A). The tumor mass showed solid areas intermingled with ductal-like areas, between variable amounts of extracellular matrix and some fibro vascular septa (Fig. 2B). The cells showed cuboidal, plasmacytoid, clear and epithelioid morphology (Fig. 2C). In the microscopic sample were observed microcysts with eosinophilic matrix inside, looser areas of adipose and chondroid appearance, and cellular diversity, but without presence of atypia or mitosis (Fig. 2D). There was no evidence of malignancy. The diagnosis was Pleomorphic Adenoma and the patient was referred to the Maxillofacial Service of the Public Hospital. The lesion was completely removed and no recurrence was observed in two years of follow-up.



Fig. 1. Photograph showing the clinical appearance of the lesion.

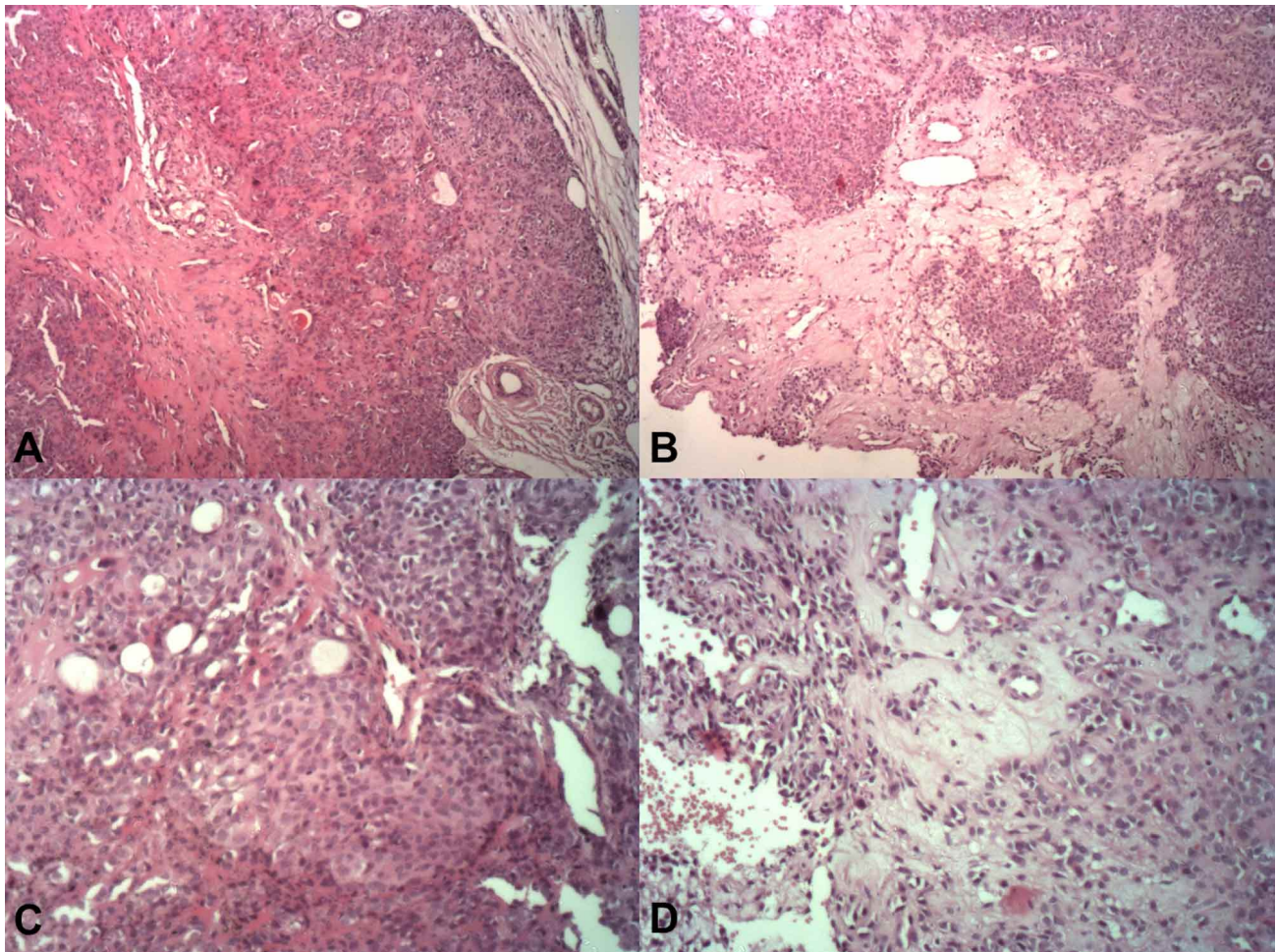


Fig. 2. Microscopic photographs showing the main features of the lesion. A) A solid lesion with hypercellular areas surrounding a fibrous central area. Note the fibrous capsule (HE, low magnification, 50X). B) Variable amount of extracellular matrix intermingled with the cellular areas (HE, low magnification, 50X). C) Some areas shows a duct-like conformation with epithelioid and plasmacytoid morphology of the cells (HE, high magnification, 100X). D) Clear cell areas showing the myoepithelial component of the lesion (HE, high magnification, 100X).

Table I. List of reported cases of Pleomorphic Adenoma in children and adolescents.

Author	Sex	Age (years)	Site
Byars <i>et al.</i> , 1957	Female	9	Hard palate
Byars <i>et al.</i> , 1957	Female	7	Hard palate
Crawford-Guempsey, 1967	Female	8	Left hard and soft palate
Galich, 1969	Female	12	Hard and soft palate
Buehrle & Friedberg, 1972	Female	13	Right hard and soft palate
Budnick, 1982	Female	12	Hard and soft palate
Budnick, 1982	Female	12	Hard palate
McIlveen <i>et al.</i> , 1987	Female	7	Hard palate
Lack and Upton, 1988	Male	10	Soft palate
Fonseca <i>et al.</i> , 1991	Female	16	Soft palate
Fonseca <i>et al.</i> , 1991	Female	8	Soft palate
Austin & Crockett, 1992	Female	10	Hard palate
Noghreyan <i>et al.</i> , 1995	Female	8	Hard palate
Lopez-Cedrun <i>et al.</i> , 1996	Male	16	Hard palate
de Courten <i>et al.</i> , 1996	Female	10	Hard palate
Chen <i>et al.</i> , 1998	Female	15	Right hard palate
Shaaban <i>et al.</i> , 2001	Male	9	Hard palate
Jorge <i>et al.</i> , 2002	Male	11	Soft palate
Jorge <i>et al.</i> , 2002	Female	17	Hard palate
Daniels <i>et al.</i> , 2007	Male	5	Left hard palate
Daniels <i>et al.</i> , 2007	Male	16	Left hard palate
Dhanuthai <i>et al.</i> , 2009	Female	13	Left hard palate
Arcuri <i>et al.</i> , 2011	Male	11	Right hard palate
Ritwik & Brannon, 2012	Female	15	Right hard palate
Ritwik & Brannon, 2012	Female	17	Left hard palate
Ritwik & Brannon, 2012	Male	12	Hard palate
Ritwik & Brannon, 2012	Female	15	Soft palate
Ritwik & Brannon, 2012	Male	12	Soft palate
Thangaswamy <i>et al.</i> , 2012	Male	19	Hard palate
Zainab, 2013	Female	13	Right hard Palate
Krishna, 2013	Female	14	Hard palate
Maclsaac <i>et al.</i> , 2013	Female	7	Right hard palate
Bovino <i>et al.</i> , 2013	Male	12	Hard palate
Hughes <i>et al.</i> , 2015	Female	ND	Soft Palate
Swain <i>et al.</i> , 2016	Male	13	Left soft palate
Current case	Female	15	Right hard palate

## DISCUSSION

Salivary gland tumors are uncommon in the first decades of life. The majority of these tumors are benign, but in minor salivary glands the incidence of malignant tumors is approximately 50 % of the reported cases. In children, major salivary glands are more affected by salivary gland tumors than minor salivary glands. PA is the most common benign lesion of minor salivary glands in children and adolescents, being the palate one of the most frequently affected site of minor salivary glands. To the best of our knowledge, 36 cases of palatal PA were reported in the first and second decade of life (Table I). The age range was 5 to 19

years-old, with a mean age of 12.4 years-old. The incidence is higher in the second decade, with 25.7% of the tumors occurring in patients younger than 10 years (Byars *et al.*, 1957; Crawford & Guernsey, 1967; Galich, 1969; Buehrle & Friedberg, 1972; Budnick, 1982; McIlveen *et al.*, 1987; Lack & Upton, 1988; Fonseca *et al.*, 1991; Austin & Crockett, 1992; Noghreyan *et al.*, 1995; Lopez-Cedrún *et al.*, 1996; de Courten *et al.*, 1996; Chen *et al.*, 1998; Shaaban *et al.*, 2001; Jorge *et al.*; Daniels *et al.*, 2007; Dhanuthai *et al.*, 2009; Arcuri *et al.*, 2011; Ritwik & Brannon, 2012; Thangaswamy *et al.*, 2012; Bovino *et al.*, 2013;

Maclsaac *et al.*, 2013; Pramod Krishna, 2013; Zainab, 2013; Hughes *et al.*, 2015; Swain *et al.*, 2016).

Byars *et al.*, reported the first 2 cases of palatal PA in children aged 7 years and 9 years (Byars *et al.*). The current case occurred in a young girl, and reports in the English literature revealed a female sex predilection (3:1). The majority was described as painless and as firm sub-mucosal masses or nodules (Byars *et al.*; Crawford & Guernsey; Galich; Buehrle & Friedberg; Budnick; McIlveen *et al.*; Lack & Upton; Fonseca *et al.*; Austin & Crockett; Noghreyan *et al.*; Lopez-Cedrún *et al.*; de Courten *et al.*; Chen *et al.*; Shaaban *et al.*; Jorge *et al.*; Daniels *et al.*; Dhanuthai *et al.*; Arcuri *et al.*; Ritwik & Brannon; Thangaswamy *et al.*; Bovino *et al.*; Maclsaac *et al.*; Pramod Krishna; Zainab; Hughes *et al.*; Swain *et al.*). Six of these cases caused underlying-bone involvement, described as pressure erosion, smooth depression, or reabsorption (Chen *et al.*; Dhanuthai *et al.*; Arcuri *et al.*; Ritwik & Brannon); and one of these cases perforated the palatal bone, extending into nasal cavity (Arcuri *et al.*). The time of evolution is usually fast-growing, being faster than in adult patients (Chen *et al.*).

Biopsy is necessary to diagnose asymptomatic palatal swelling of firm consistency. Histopathologically, PA is composed of a wide spectrum of epithelial and mesenchymal tissue derived from cells with ductal and myoepithelial features with a rich stroma, often with myxomatous appearance (Chen *et al.*; Jorge *et al.*; Dhanuthai *et al.*; Arcuri *et al.*; Ritwik & Brannon), and some cases with the presence of cartilaginous or osteoid formation inside the tumor (Shaaban *et al.*). The treatment of PA is the same in adults, and the recurrence of PAs in minor salivary glands of children was rarely reported, but we do not know if these recurrences were associated with the tumor behavior or surgical problems (Daniels *et al.*). Recurrence on the palate could be a serious complication, because is necessary greater margins of the surgery.

In conclusion, PA rarely occurs before the second decade of life, but is the most common salivary gland neoplasm in children and adolescents, with a female predilection, and should be considered in the differential diagnosis of young patients with swellings in the palate. This case report will contribute to a better characterization of PA in children and adolescents, determining a true sex predilection and age of distribution with a higher number of reported cases.

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**RESUMEN:** El adenoma pleomórfico (AP) es la neoplasia benigna más común de las glándulas salivales y se diagnostica frecuentemente en la tercera y cuarta década con predilección por las mujeres. El AP es la lesión benigna más común de las glándulas salivales menores en niños y adolescentes, siendo el paladar uno de los sitios más frecuentemente afectados de las glándulas salivales menores. En este trabajo se presenta un relato de caso de un AP de paladar duro diagnosticado en una mujer de 15 años de edad y una revisión de la literatura en inglés de los casos reportados de AP en niños y adolescentes en paladar duro.

**PALABRAS CLAVE:** boca, hemangioma, malformación vascular.

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