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.

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
x 6 mm of size. The histopathological examination noted a solid lesion covered by an orthokeratinized pluristratified 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.
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., 2002; Daniels et al., 2007; Dhanuthai et al., 2009; Arcuri et al., 2011; Ritwik & Brannon, 2012; Thangaswamy et al., 2012; Bovino et al., 2013;
MacIsaac 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; Mclliveen et al.; Lack & Upton; Fonseca et al.; Austin & Crockett; Noghreian 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.; MacIsaac 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.

REFERENCES


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.


