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García, Pedro Emilio; Avila, Rodolfo Esteban; Samar, María Elena
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Case report of Basal cell adenocarcinoma of the parotid gland: clinicopathological and immunohistochemical study

García Pedro Emilio¹, Avila Rodolfo Esteban², Samar María Elena³

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Abstract

Basal cell adenocarcinoma is an epithelial neoplasm with the cytological characteristics of basal cell adenoma but with a morphological pattern of infiltrative growth indicative of malignancy. Due to its low incidence it is often difficult to diagnose a basal cell adenocarcinoma. The objective of the present study was to identify morphological and immunohistochemical characteristics that contribute to its diagnosis. A parotid tumor was resected in a 52-year-old patient; postoperative biopsy and immunostaining with Ki-67, CK19, p63 and alpha- smooth muscle actin were performed. It was diagnosed basal cell adenocarcinoma that invades the tumor capsule, periglandular fat and lymph nodes. Immunostaining with Ki-67, CK19, p63 and alpha- smooth muscle actin was positive. Subsequently, a maxillary sinus metastasis was diagnosed. The morphological characteristics, Ki-67 expression strongly positive and metastasis give the malignant character to this tumor, which differentiates it from the basal cell adenoma.

Keywords: parotid, basal cell adenocarcinoma, diagnosis.

- 1 Medical Doctor - Clinical Oncology Specialist. Assistant Professor. School of Medical Sciences. Universidad Nacional de Córdoba. Argentina. ORCID: 0000-0001-5268-2339
- 2 Doctor of Medicine and Surgery. Associate Professor. School of Medical Sciences. Universidad Nacional de Córdoba. Argentina. ORCID: 0000-0001-8857-2406
- 3 Doctor of Medicine and Surgery. Full Professor. School of Dentistry. Universidad Nacional de Córdoba. Argentina. ORCID: 0000-0002-6093-3297

Introduction

The World Health Organization as well as Ellis and Auclair described basal cell adenocarcinoma (ICD-O Code 8147/3, International Classification of Diseases for Oncology) as a cytological epithelial neoplasm, and in terms of histomorphology it was found to be very similar to basal cell adenoma, but with infiltrative growth and a low incidence of metastasis⁽¹⁻³⁾.

Background

Some authors consider that this tumor stems from a basal cell adenoma while others argue that it originates *de novo*. Of all the cases, 80% are located in the parotid glands, 9% in the submandibular gland and 11% in minor salivary glands. It is a rare tumor, which is more frequent in adults between their 4th and 9th decade of life; it is very rare in children. Swelling is usually the only symptom but sometimes it also causes pain and occasionally remains undiagnosed for ten years. It has been categorized into four types based on its histological pattern of growth: solid, membranous, trabecular and tubular⁽⁴⁻⁶⁾. Given its low incidence and scarce bibliographic information, it is often difficult to diagnose it⁽⁷⁾.

The aim of this study was to present a case of basal cell adenocarcinoma in the parotid deep lobe in a 52-year-old woman and to identify morphological and immunohistochemical characteristics that contribute to its diagnosis. This work is part of the project called "The extracellular matrix and the myoepithelial component of epithelial tumors of human salivary glands: structural, histochemical, immunohistochemical and lectin histochemical study", authorized by the Ethics Committee of the National Clinical Hospital. School of Medical Sciences. Universidad Nacional de Córdoba. Argentina (File 188/14).

Clinical case

In November 2002, a 52-year-old female patient visited the dentist and an otorhinolaryngology service since she felt pain in the left region of the mandible; a diagnosis was not reached. In December 2002 she went to a new appointment and a nodular mass was found in the left ascending ramus of the mandible, with increased pain on palpation. She was diagnosed with trigeminal neuralgia, ruling out the importance of the nodular lesion; physiotherapy was indicated for the cervical region. However, the pain continued to increase, so in December 2003 she underwent a biopsy puncture of an adenopathy, where an inflammatory process was identified.

In January 2004, a medical interconsultation was made with a head and neck surgeon who requested a CT scan of the neck which showed an image compatible with a tumor lesion. In February of the same year, a left parotid deep lobe tumor was excised with good postoperative evolution.

Anatomic pathology report: The macroscopy showed a slightly lobed, encapsulated mass, 5 cm x 1.7 cm x 1.2 cm of violet gray color with firm areas when cut. In the microscopic examination of the colored sections with H/E there was a fibrous capsule that surrounded the basaloid cells with pale eosinophilic cytoplasm and rounded or oval nuclei organized in sheets and nests of variable shapes and sizes, separated by bands of connective stroma. No areas of tumor necrosis, cellular atypia or mitotic figures were found.

The histological growth pattern was solid and tubular. The solid pattern presented nests and strands of contiguous basaloid cells that in the periphery of these structures formed a cellular layer organized in palisade cells.

In the tubular pattern there were basaloid cell islets that contained pseudocysts or prominent lumen lined with cuboid cells (Fig. 1 A and B).

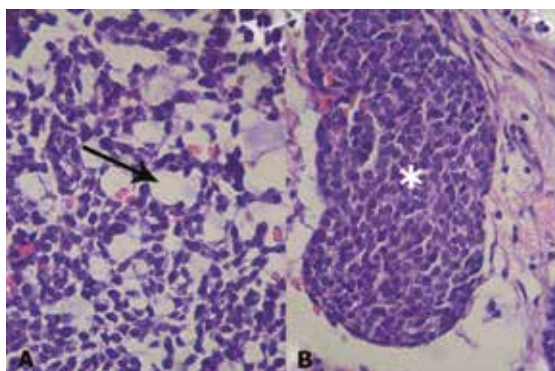


Fig. 1: HISTOLOGICAL PATTERNS OF BASAL CELL ADENOCARCINOMA

Tubular pattern. The arrow indicates the cells that contain lights and pseudocysts lined by cells with dense chromatin nuclei. H/E 400x.

Solid pattern. Nest of basaloid cells (asterisk). H/E 400x.

The diagnosis of mixed type basal cell adenocarcinoma was made on the basis of its histological patterns (solid and tubular) and its malignant character, with an aggressive growth with infiltration of the tumor capsule and periparotid fat, perineural invasion and metastasis of a regional lymphoid node (Fig. 2A, B and C).

We then observed maxillary sinus involvement with bone metastasis.

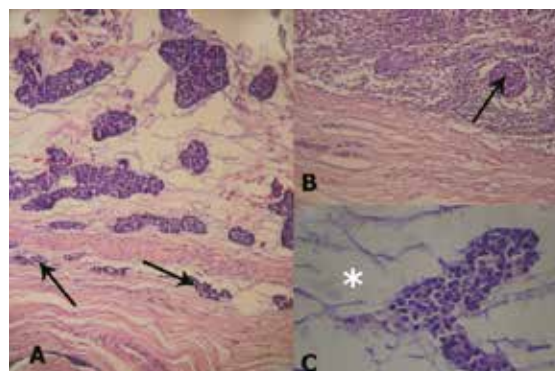


Fig. 2: BASAL CELL ADENOCARCINOMA WITH INFILTRATING GROWTH AND METASTASIS

The arrows show the invasion of the tumoral connective capsule. H/E 100x.

The arrow shows the metastasis in a regional lymphoid node (H/E). 100x.

The asterisk shows the infiltration of periparotid adipose tissue. H/E 400x.

Immunohistochemical labeling was used to find the differentiation of basaloid cells in epithelial (CK 19) and myoepithelial cells (p63 and α -smooth muscle actin) (Fig. 3A). The proliferating capacity of the tumor was studied with the marker for Ki-67. A positive score was given only if there were over 10% of marked cells⁽⁸⁾: the result was 25% of positive cells. (Fig. 3B). The results are shown in Table 1.

Table 1: Immunohistochemical analysis of basal cell adenocarcinoma

Marker	Marked cell	Reaction *
Ki- 67 (prognostic marker of cell proliferation)	Epithelial cells (nuclei)	Strong
p63 (myogenic differentiation)	Myoepithelial cells (nuclei)	Strong
Alpha-AML (myogenic differentiation)	Myoepithelial cells (cytoplasm)	Moderate
CK19 (epithelial differentiation)	Epithelial cells (cytoplasm)	Strong

Alpha-AML: α -smooth muscle actin. CK19: cytokeratin 19

* The qualitative assessment of the labeling was performed according to the intensity of coloration.

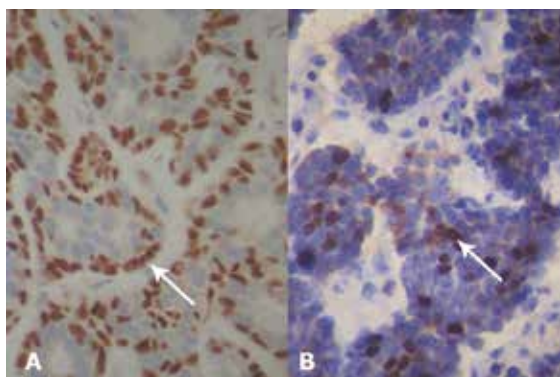


Fig. 3: IMMUNOHISTOCHEMICAL LABELING OF BASAL CELL ADENOCARCINOMA

The arrow shows the nuclei of myoepithelial cells positive for p63. 400x.

The arrow indicates the nuclei of the epithelial cells positively labeled for Ki-67. 400x.

Given the results of the histopathological study, the patient was referred for oncological treatment, with indication of postoperative radiotherapy for six weeks, which began in March 2004. The treatment plan included the irradiation of the parotid plus the cervical region left at a total dose of 60 Gy, with a daily dose of 2 Gy. The complications were mucositis, dysphagia and mycosis, which, upon receiving specific treatment, improved at the end of the radiotherapy sessions.

In September 2004, an orthopantomography showed a cyst in the left horizontal ramus of the mandible and a vegetative image in the homolateral piriform sinus. The surgery was performed and the biopsy was negative for malignancy.

Subsequently, in March 2005, the pain in the hard palate and the left region of the maxilla began. A CAT scan was indicated and a lesion was identified at the maxillary floor level. The surgery was performed and metastasis was diagnosed in the floor of the maxillary sinus. A new radiotherapy plan was indicated, which had mucositis and dysphagia as complications. The general condition of the patient worsened and she presented dehydration and drowsiness.

The radiation therapy scheme could not be completed; palliative care was continued until her death two months later.

Discussion

Primary basaloid tumors of the salivary glands are lesions characterized by a predominance of “basaloid” epithelial cells with round or ovoid nuclei surrounded by a thin cytoplasm. The term “basaloid epithelial cells” arose from observations with standard histological techniques that do not make it possible to distinguish if there are myoepithelial or ductal or basal epithelial cells. In these cases, immunohistochemical labeling is a useful tool to detect both cell types, since it improves diagnostic accuracy. The immunohistochemistry panel used in the case described (p63, α -smooth muscle actin and CK19) was important for the diagnosis of the “basaloid” cells of this carcinoma^(9,10).

The heterogeneous tumoral group of basaloid epithelial cells includes benign tumors such as pleomorphic adenoma, and basal cell adenoma, and malignant tumors such as adenoid cystic carcinoma, undifferentiated small cell carcinoma, and basal cell adenocarcinoma^(10,11).

Basal cell adenocarcinoma mainly affects the parotid gland (more than 90%) and is histologically similar to basal cell adenoma. However, it presents an infiltrating growth and a low incidence of metastasis^(1,2,12).

It is generally considered a carcinoma of low degree of malignancy, locally destructive and with frequent recurrences. The recurrence period varies from six months to two years and only occasionally produces distant metastases^(12,13).

In this case the evolution was atypical, presented a bone metastasis in an early form, less than a year after the end of its treatment. This led to the deterioration of the general condition of the patient and her death. This can be explained by the late diagnosis of the lesion, with lymphoid node metastasis; and histological factors of

poor prognosis such as capsular, perineural and periglandular fat invasion.

There is no predilection for gender, and 80% of tumors occur after 50 years of age with an average of 60 years of age⁽¹⁴⁾. This case corresponds to a 52-year-old woman.

Its histomorphological patterns are described as solid, membranous, tubular or trabecular. The solid pattern is the most frequent, characterized by solid cellular nests of different shapes and sizes. A collagen stroma, consisting of bands of different thickness, separates these nests that correspond to the most frequent histological pattern and with the highest risk of metastasis. In the membranous pattern there are abundant deposits of a strongly eosinophilic hyalinized basal membrane. The growth pattern is tubular when luminal spaces are formed between basaloid epithelial cells, whereas in trabecular growth, basaloid epithelial cells are arranged in interconnected cell bands⁽¹⁵⁾.

In our study, the tumor was located in the parotid gland with a mixed growth pattern since the basaloid epithelial cells were placed in solid and tubular areas.

Regarding the prevalence of this neoplasia in Latin America, in the consulted databases we only found data in two studies by Ruiz-Godoy and Rivera et al. and Ito et al.^(16,17).

In 1996 Ruiz-Godoy Rivera et al. conducted a clinicopathological study of six cases of basal cell adenocarcinoma analyzed at the Instituto Nacional de Cancerología, México⁽¹⁶⁾.

Ito et al. examined the archives of the Department of Pathology of the Instituto del Câncer de Londrina (Paraná State, Brazil) for the years 1972-2001, and analyzed a total of 496 cases of major and minor salivary gland tumors. These researchers found only three cases of basal cell adenocarcinoma⁽¹⁷⁾.

Histologically, the main differential diagnosis is made with basal cell adenoma; but it also includes the solid variant of cystic adenoid carcinoma, basaloid squamous cell carcinoma and

cutaneous basal cell carcinoma with deep invasion^(12,15).

Wilson and Robins claim that local invasion of the surrounding soft tissues and the gland are the best markers for differentiating the basal cell adenoma from basal cell adenocarcinoma. They also consider it important to complement the recognition of these tumors on the bases of invasion with proliferation markers such as Ki-67 and apoptosis markers⁽¹⁸⁾.

In a previous study we obtained a negative Ki-67 labeling in the basal cell adenoma, unlike the basal cell adenocarcinoma, where we observed a proliferation index greater than 25%, which also explains the unfavorable evolution of this patient⁽¹⁰⁾.

Saluja et al. also claim that the definitive diagnosis of malignancy is established by the invasive nature of the neoplasm in the surrounding tissues⁽¹⁹⁾.

On the other hand, Jung et al. report that basal cell adenoma with capsular invasion can also be considered an infiltrating tumor, which makes us question the category of basal cell adenocarcinoma. These authors say that the tumors do not have morphological differences, frequently develop a cribriform or solid growth pattern and are larger than the basal cell adenoma without capsular invasion. However, they consider that the cribriform structure of basal cell adenocarcinoma may correspond to a misdiagnosis of adenoid cystic carcinoma. In addition, the solid variant of adenoid cystic carcinoma presents cells with marked nuclear atypia and frequent mitosis⁽⁷⁾.

Although regional and distant metastases are not common, in this case there was metastasis in a regional lymphoid node and also in the maxillary sinus floor, which led to the patient's death⁽¹²⁾.

In general, there is no consensus regarding its treatment, since some recommend local excision and others total parotidectomy, even in cases of membranous basal cell adenoma, as mentioned by Zhan et al.⁽¹²⁾.

For the primary treatment of the tumor, when it originates in the minor salivary glands, some authors consider that surgical excision should be wide to ensure its complete removal⁽²⁰⁾.

In addition, there is no agreement on the role of radiotherapy. However, postoperative radiotherapy is recommended in the case of surgeries with positive surgical margins or surgical excision of recurrent tumors^(21,22).

Conclusions

Epithelial and myoepithelial cells coexist in the basal cell adenocarcinoma of the salivary glands, identified through immunohistochemical markers.

It is essential to obtain histological sections of the tumor interface with the normal gland or with the surrounding fat to evaluate tumor invasion.

Capsular, perineural and periparotid adipose tissue invasion, Ki-67 positive immunolabeling with a proliferative index of 25% and metastasis in a regional lymphoid node and in the maxillary sinus are factors of poor prognosis that, associated with late diagnosis, explain the unfavorable evolution of the patient.

Although it is a tumor with local aggressiveness and low incidence of metastasis, early diagnosis and early oncological care are of crucial importance to give these patients a greater chance for a cure.

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María Elena Samar: samarcongreso@gmail.com

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