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Basal Cell Adenoma: A Case Report

ANA ROSA FERRETE ESTEVES, DDS,* LUCIANO LAURIA DIB, DDS, PHD,† AND LEDA VIEGAS DE CARVALHO, MD‡

Basal cell adenoma is a rare benign neoplasm of the salivary glands that derives its name from the basaloid appearance of the tumor cells. It preferentially occurs in the parotid gland and upper lip during the sixth and seventh decades of life.[1] The clinical presentation most frequently seen is a slow-growing, asymptomatic, movable, round or oval, normally colored submucosal mass measuring less than 3.0 cm in diameter, encapsulated and well circumscribed.[1,3] Histologically, the tumor consists of a proliferation of the terminal duct epithelial cells forming islands[4] and sheets supported by a sparsely fibrous stroma, and the presence of small numbers of myoepithelial cells.[1]

Basal cell adenoma is an uncommon tumor,[4] and a case involving the palate has not been previously reported.

Report of Case

A 36-year-old black woman was referred to the Stomatology Department of A.C. Camargo Hospital in May 1994 complaining of a swelling on the hard palate of 4 months' duration. The patient reported that she had had an incisional biopsy at another institution one month before. Microscopic examination led to a diagnosis of basal cell adenoma.

Intraoral examination showed a movable, firm, painful, submucosal mass covered with reddish mucosa, measuring 2.0 cm in the widest diameter and located on the right side of the hard palate. The surface was ulcerated (Fig 1).

Radiographic examination with panoramic and occlusal views, as well as a computed tomography (CT) scan, showed no signs of bone destruction. However, the lesion had caused slight elevation of the floor of the right nasal cavity.

The patient was treated under general anesthesia by complete excision of the lesion, including a small security margin (Fig 2). Microscopic examination showed multiple islands and groups of epithelial cells with ductlike structures supported by a small amount of fibrous stroma (Fig 3). The hyperchromatic peripheral cells of these islands were palisaded and cuboidal to columnar in shape (Figs 4, 5). The central cells of the islands tended to have paler staining nuclei and, occasionally, formed eddies or keratin pearls. Alternating with the epithelial sheets were ductlike structures, characterizing the lesion as a trabecular-tubular subtype.

The postoperative cause was uneventful, and after a follow-up of 16 months, there are no signs of local recurrence (Fig 6).

Discussion

The basal cell adenoma was once considered to be a type of "monomorphic adenoma."[4] However, since 1991, according to the "Salivary Glands Tumours Histological Classification" of the World Health Organization, the name of this lesion was changed to basal cell adenoma, excluding the word "monomorphic."[5]

Among the "monomorphic adenomas," there are the following varieties: Warthin's tumor or papillary cystadenoma lymphomatosum, oncocytoma or oxyphilic adenoma, basal cell adenoma, canalicular adenoma, and sebaceous adenoma.[1,6] Histogenetically, they can be divided into four groups: 1) tumors of terminal duct origin (basal cell adenoma and canalicu-

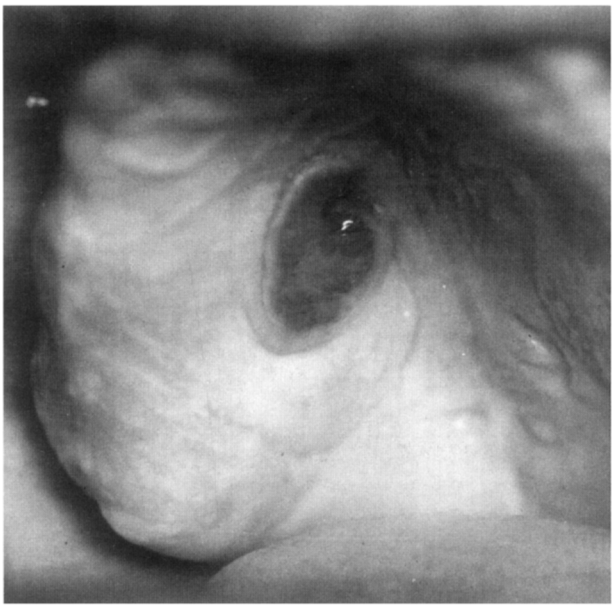

FIGURE 1. Intraoral view showing ulcerative lesion involving right hard palate

Received from the A.C. Camargo Hospital, São Paulo, Brazil.
* Senior Resident, Stomatology Department.
† Chairman, Stomatology Department.
‡ Pathologist, Pathology Department.
Address correspondence and reprint requests to Dr Dib: Departamento de Estomatologia, Hospital A.C. Camargo, Rua Professor Antônio Prudente, 211, Liberdade—São Paulo-SP 01509-010.

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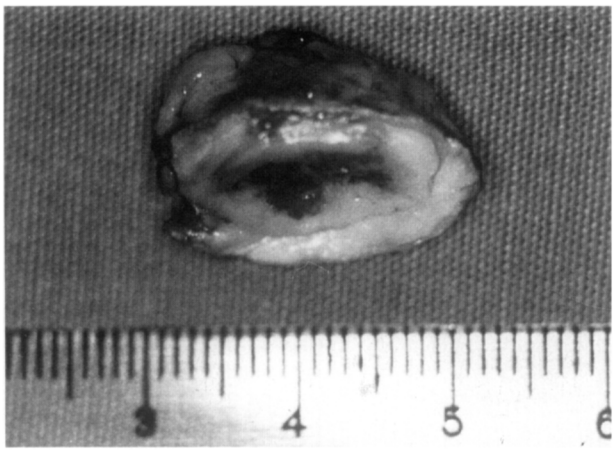

FIGURE 2. View of resected specimen.

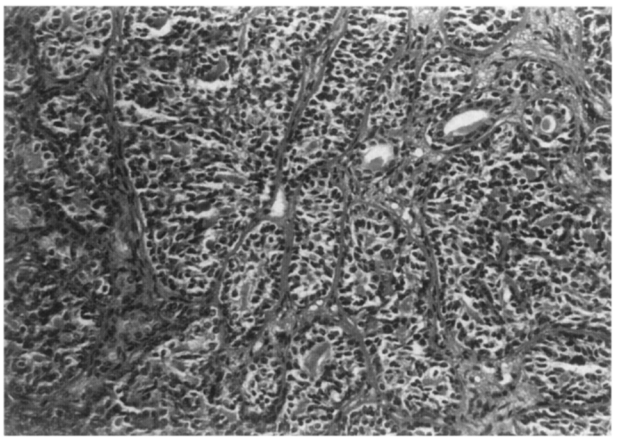

FIGURE 4. High-power view showing sharp demarcation between islands of neoplastic epithelial cells and peripheral palisading (hematoxylin-eosin stain; original magnification ×160).

lar adenoma), 2) tumors of terminal or striated duct origin (sebaceous adenoma and sebaceous lymphadenoma), 3) tumors of striated duct origin (oncocytoma and papillary cystadenoma lymphomatosum), and 4) tumors of excretory duct origin (sialadenoma papilliferum or inverted ductal papilloma).[6,7]

The salivary gland tumors are uncommon, representing less than 3% of all neoplasms of the head and neck.[8] Although it is the most common variant in the group of "monomorphic adenomas," basal cell adenoma represents only 1% of all salivary tumors.[1]

The literature is controversial about sex predominance. Some authors have reported male[6] predominance and others female predominance.[1,4] The tumor can occur at any age but is most common in middle-aged and older adults, with a peak prevalence in the seventh decade of life.[1,9,10] In our case, the patient was in the fourth decade of life, in contrast to the literature data.

The basal cell adenoma can occur in all salivary tissues but is more frequent in the parotid gland,[9] followed by the minor salivary glands of the upper lip.[1,11] The development of these tumors in the buccal mucosa, palate, or lower lip is unusual.[4,12] In the current case, the palatal location of the tumor did not fit the more frequent sites, and the literature reviewed showed no reports of occurrence in this region.

The origin of the basal cell adenoma is epithelial, probably in the cells of the terminal duct. Frequently, a mixture of histopathologic subtypes is seen, that is, tubular areas alternating with trabecular and solid areas.[1]

Among the malignant tumors, the adenoid cystic carcinoma[11] is the lesion that shows the most histologic similarities to the basal cell adenoma, suggesting that the latter is the benign homologue of the adenoid cystic carcinoma. However, characteristics such as integrity

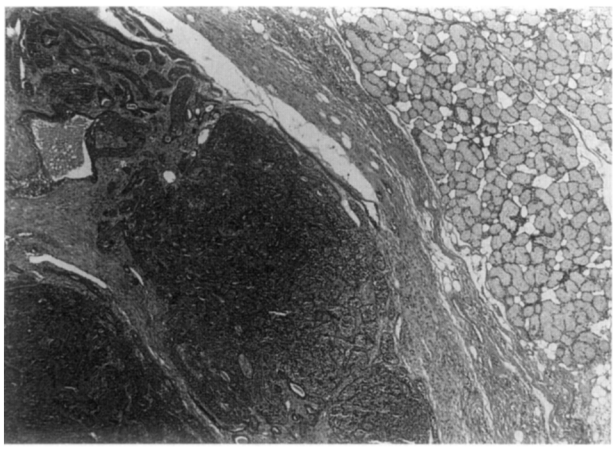

FIGURE 3. Low-power view showing well-circumscribed and encapsulated basal cell adenoma (hematoxylin-eosin stain, original magnification ×40).

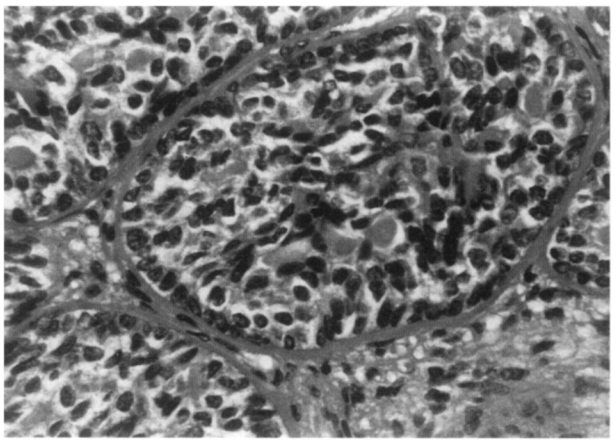

FIGURE 5. High-power view showing basal cell adenoma with a tubular pattern (Hematoxylin-eosin stain; original magnification ×400).

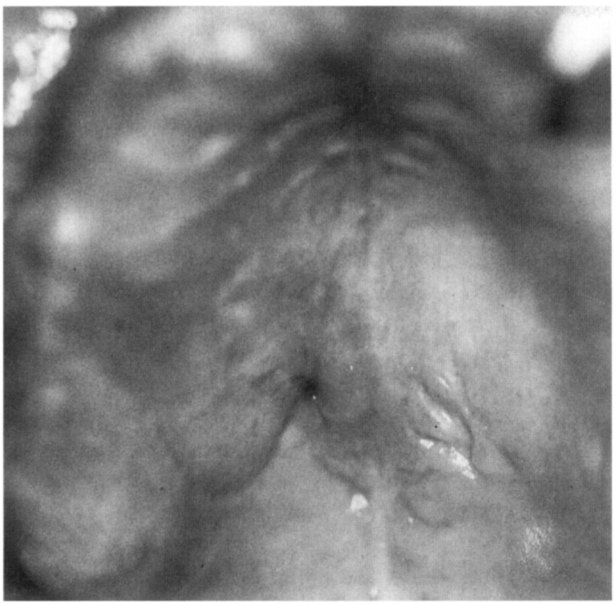

FIGURE 6. Postoperative view showing healing of right hard palate with a slight mucosal defect.

of the basal layer, decreased number of mitoses, and slow growth are typical of a benign lesion.

The basal cell adenocarcinoma is another malignant tumor that shares histologic features with the basal cell adenoma. Both exhibit myoepithelial differentiation, reactivity patterns indicative of ductal epithelium, and closely similar immunohistochemical profiles. Basal cell adenocarcinoma is distinguished from basal cell adenoma by the histologic features of invasion, mitotic activity, and neural or vascular involvement.[13-15]

The differential diagnosis must include the pleomorphic adenoma, which is the most common benign tumor of the salivary glands, and other salivary gland tumors such as the canalicular adenoma and sebaceous adenoma. Malignant tumors must be ruled out, and the clinical aspects such as time of evolution, ulceration, and bone infiltration should be well evaluated even though none of these characteristics are a pathognomonic signal of malignancy.[12] In the case reported, the lesion showed ulceration, but this condition was attributed to the previous incisional biopsy.

Malignant transformation to basal cell adenocarcinoma is rare[6] but is suggested by some authors. Although recurrence is rare, the membranous subtype, which is a hereditary variety of basal cell adenoma, has a 25% to 37% recurrence rate, possibly related to its multifocal nature, which impairs complete removal.[1,4,10]

The membranous subtype shows association with skin adnexal tumors (dermal cylindromas and trichoepitheliomas[1,4]). Batsakis et al have used the term dermal analogue tumors to describe membranous "monomorphic adenomas."[16] Histologically, the epithelium is typical of the basal cell type adenoma, the only distinguishing feature being the thick hyaline sheaths surrounding the epithelium.[4]

The treatment used in this case was the same proposed in the literature,[1,4,11] consisting of complete surgical removal with an extracapsular limit.[1] The patient had a satisfactory postoperative period, with complete healing of the operated area, and presents no signs of local recurrence 16 months after surgery.

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