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Hybrid tumours of the salivary glands. A report of two cases involving the palate and a review of the literature

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Abstract Hybrid tumours are very rare salivary gland lesions composed of two or more different tumoural entities in a single neoplasm that arise within a definite topographical region. In most cases adenoid cystic carcinoma has been the predominant component in these lesions. In this study we describe two patients with hybrid tumours located in the palate, one in a 49-year-old woman and one in a 71-year-old man. The first case involved adenoid cystic carcinoma and mucoepidermoid carcinoma, and the patient in the second case exhibited adenoid cystic carcinoma and epithelial-myoepithelial carcinoma. Both patients were treated with surgery and radiotherapy, and there has been no evidence of recurrence after 13 and 36 months of follow-up, respectively. The recognition of the histologic component with the higher grade of malignancy in every case of hybrid tumour of the salivary glands is a necessary step to determine the biological behaviour and, consequently, to determine the proper therapeutic approach.

Keywords Hybrid tumours · Salivary glands · Carcinoma

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Introduction

The classification of salivary gland tumours includes a large number of lesions, some of which exhibit a wide range of histopathologic variants. Recently, Seifert et al. [10] described five cases of "very rare tumoural entities, each of which conforms with an exactly defined tumour category" and designated them as hybrid tumours. To date, less than twenty cases have been published in the English literature [1, 3, 4, 6, 7, 10, 12, 14, 15].

The purpose of this article is to present two new cases of hybrid tumours of the salivary glands, focusing on their histomorphological features, as well as comparing them with previously reported cases of these unusual lesions.

Case report

Case 1

A 49-year-old woman complained of a painless palatine tumour that had been growing slowly for 10 years, but in the last 8 months, it had grown more quickly and become ulcerated. On clinical examination an exophytic, multilobulated and ulcerated mass involving both the right hard and soft palate was evident. The lesion measured 3.5 cm and extended beyond the midline. A neck examination disclosed no lymphadenopathy. Treatment consisted of maxillectomy and postoperative radiotherapy (60 Gy). Thirteen months later the patient is alive with no evidence of recurrence.

Gross examination disclosed a 3.5×3.5×2.5 cm ulcerated exophytic neoplastic growth located along the hard palate, which extended back to the soft palate and infiltrated beyond the midline. Macroscopically, the tumour was yellowish white, firm and partially solid, with small cystic spaces scattered in the lesional tissue.

Microscopic examination disclosed two morphological patterns: adenoid cystic carcinoma and mucoepidermoid carcinoma. The first was composed of a predominantly solid pattern with occasional areas of cribriform morphology. The mucoepidermoid component showed a cystic pattern with abundant mucous-producing cells and a moderate amount of intermediate and epidermoid cells (Fig. 1a and Fig. 1b).

Case 2

The patient was a 71-year-old man who presented with a 7-month history of a tumour mass located in the right hard and soft palate.

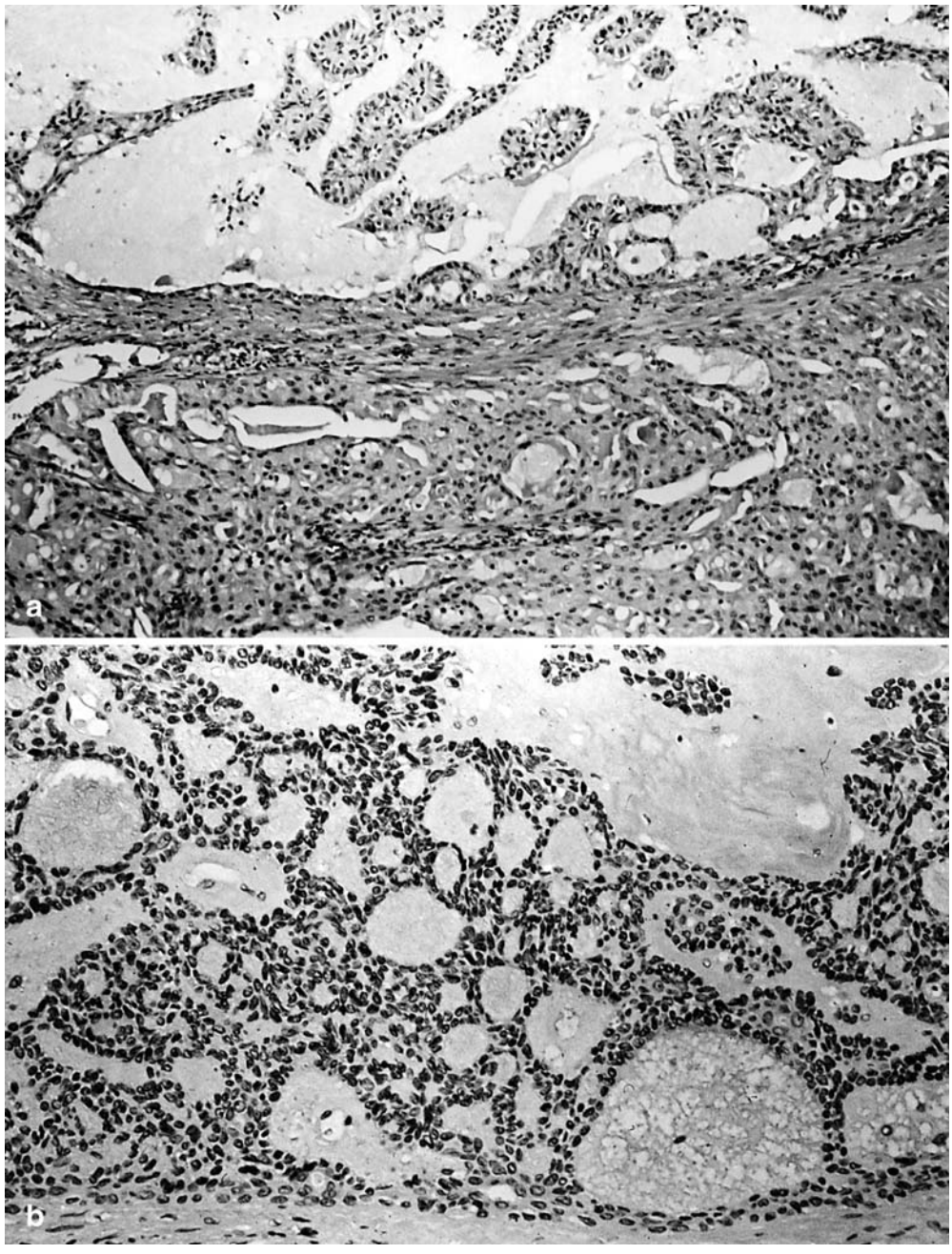

Fig. 1 **a** Case one shows a mucoepidermoid carcinoma component (HE, ×250). **b** Case one shows an area of adenoid cystic carcinoma with a cribriform pattern (HE, ×250)

The lesion was firm and tender. The patient also had associated facial paralysis. A CT scan disclosed a tumoural mass located within the right nasal fossa and maxillary sinus, which produced deviation of the nasal septum and thickening of the anterior ethmoidal cells. Treatment consisted in a right maxillectomy and postoperative radiotherapy of 60 Gy. After 36 months of follow-up, there has been no evidence of recurrent disease.

Macroscopically, the surgical specimen showed a tumour located in the hard and soft palate that infiltrated the maxillary sinus. The tumour was solid, yellowish and firm in consistency with some cystic areas.

Microscopic examination disclosed two different morphological patterns: adenoid cystic carcinoma and epithelial-myoepithelial carcinoma. The first one exhibited tubular and cribriform patterns, while the epithelial-myoepithelial carcinoma showed the classic pattern, with ductal cells surrounded by myoepithelial cells (Fig. 2a and Fig. 2b).

Discussion

Hybrid tumours are very rare salivary gland neoplasms composed of two or more different tumoural entities in a single neoplasm that arise within a definite topographical region [10]. It is important to differentiate these neoplasms from collision tumours, which are lesions that originate in different regions but coalesce in a particular area [3]. Also, these lesions should be distinguished from tumours with biphasic differentiation, which are single entities composed of two different cellular types, such as epithelial-myoepithelial carcinomas and pleomorphic adenomas, for example [6, 7, 12].

It is interesting to note that most of the previously published cases of hybrid tumours of the salivary glands have

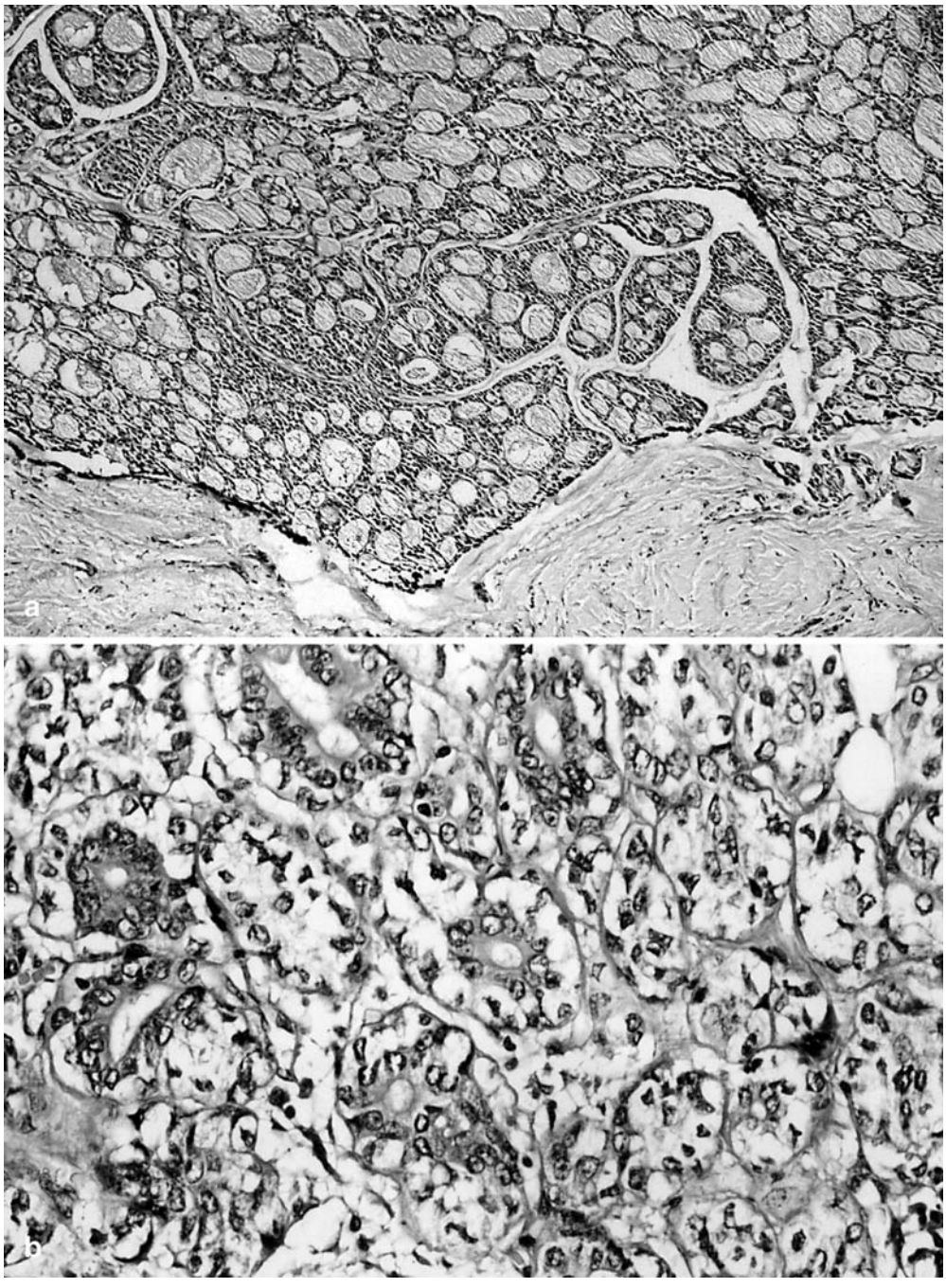

Fig. 2 **a** Case two shows adenoid cystic carcinoma with a cribriform component (HE, ×150). **b** Case two with epithelial-myoepithelial carcinoma showing ductal central differentiation surrounded by clear cells (HE, ×250)

shown a higher incidence of adenoid cystic carcinoma, which is also evident in the present series. The first case described in this article presents a mixture of adenoid cystic carcinoma plus mucoepidermoid carcinoma, and the tumour in the second was composed of adenoid cystic carcinoma and epithelial-myoepithelial carcinoma. These findings suggest that the frequent occurrence of hybrid tumours associated with this particular type of adenocarcinoma could be higher than has been estimated (Table 1).

In those cases where both cellular components are found in biphasic neoplasms, their origin may be explained by a single precursor cell that gives rise to both entities; however, most of the reported hybrid tumours have been composed of one biphasic entity and one that is not, which suggests that these neoplasms may be derived from salivary duct stem cells [2, 8, 9, 13].

Shikani et al. in 1993 reported two cases of Warthin's tumours associated with other neoplasms and presented a review of the literature, where they found 42 similar cases [11]. According to the histopathologic features described in their cases and the criteria stated by Seifert and Donath to diagnose a hybrid tumour, some of them could be classified as this entity [10].

The relevance of diagnosing hybrid tumours lies in the fact that the component with the higher grade of malignancy determines the biological behaviour of the neoplasm and, consequently, determines the proper management of each case.

Table 1 Hybrid tumours of salivary glands. *NED* no evidence of disease, *AWOD* alive without disease, *AWD* alive with disease

Case	Age	Sex	Site	Size (cm)	Histologic diagnosis	Follow-up	Authors
1	70	M	Parotid	7×6×4	Basal cell adenoma/canalicular adenoma	–	Seifert and Donath, 1996 [10]
2	62	M	Parotid	–	Basal cell adenoma/adenoid cystic carcinoma	–	Seifert and Donath, 1996 [10]
3	60	M	Parotid	5×2.5×3	Adenoid cystic carcinoma/Warthin's tumour/sebaceous lymphadenoma	–	Seifert and Donath, 1996 [10]
4	53	M	Parotid	6×3×2	Acinic cell carcinoma/salivary duct carcinoma	–	Seifert and Donath, 1996 [10]
5	66	F	Palate	–	Epithelial-myoepithelial carcinoma/ adenoid cystic.carcinoma	–	Seifert and Donath, 1996 [10]
6			Left palate	2.5	Epithelial myepithelial/adenoid cystic carcinoma/basal cell adenocarcinoma	–	Ellis et al., 1996 [5]
7	67	F	Parotid	5.5	Acinic cell carcinoma/mucoepidermoid carcinoma	NED at 18 months	Ballestin et al., 1996 [1]
8	51	M	Right palate	4.5×3.0	Adenoid cystic carcinoma/salivary duct carcinoma	Died at 19 months	Kamio et al., 1997 [7]
9	62	F	Parotid	3.0	Epithelial-myoepithelial/adenoid cystic carcinoma	NED at 20 months	Simpson 1997, [12]
10	53	M	Parotid	6.0×4.5×3.5	Adenoid cystic carcinoma/muco-epidermoid carcinoma	AWOD	Cloitoru et al., 1999 [3]
11	71	M	Parotid	2.9	Adenoid cystic carcinoma/epithelial-myoepithelial carcinoma	AWOD	Cloitoru et al., 1999 [3]
12	28	M	Parotid	2.5×2.0	Epithelial-myoepithelial carcinoma/ salivary duct carcinoma	AWD	Cloitoru et al., 1999 [3]
13	51	M	Left palate	3.5×3.0	Adenoid cystic carcinoma/salivary duct carcinoma	AWD	Cloitoru et al., 1999 [3]
14	36	F	Submandib-ular gland	3.5×2.5×2	Salivary duct carcinoma/adenoid cystic carcinoma	–	Syner and Paulino, 1999 [14]
15	49	F	Left palate	3.5×3.5×2.5	Mucoepidermoid carcinoma/adenoid cystic carcinoma	AWOD at 10 months	Ruíz-Godoy et al.
16	71	M	Right palate	4×3×3	Epithelial-myoepithelial carcinoma/ adenoid cystic carcinoma	AWOD at 49 months	Ruíz-Godoy et al.

References

1. Ballestein C, Lopez-Carreira M, Lopez JI (1996) Combined acinic cell and epidermoid carcinoma of the parotid gland. A report of a case with an immunochemical study. APMIS 104: 99–102
2. Batsakis JG, Regezi JA, Luna MA, El Naggar A (1989) Histogenesis of salivary gland neoplasms: a postulate with prognostic implications. J Laryngol Otol 103: 939–944
3. Chetty R (2000) Intercalated duct hyperplasia: possible relationship to epithelial-myoepithelial carcinoma and hybrid tumors of salivary glands. Histopathology 37: 260–263
4. Croitoru MC, Suarez AP, Luna MA (1999) Hybrid carcinomas of salivary glands. Report of four cases and review of the literature. Arch Pathol Lab Med 123: 698–702
5. Ellis GL, Auclair PL, Gnepp DR (eds) (1991) Surgical pathology of the salivary glands. WB Saunders, Philadelphia, pp 455–488
6. Grenko TR, Abendroth SC, Davis TA, Levin JR, Darick I (1998) Hybrid tumors or salivary gland tumors sharing common differentiation pathways? Reexamining adenoid cystic and epithelial-myoepithelial carcinomas. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 86: 188–195
7. Kamio N, Tanaka Y, Mukai M, Ikeda E, Kuramochi S, Fujii M, et al (1997) A hybrid carcinoma: adenoid cystic carcinoma and salivary duct carcinoma of the salivary gland. An immunohistochemical study. Virchows Arch 430: 495–500
8. Martins C, Fonseca I, Roque L, Ribeiro C, Soares J (2001) Cytogenetic similarities between two types of salivary gland carcinomas: adenoid cystic carcinoma and polymorphous low-grade adenocarcinoma. Cancer Genet Cytogenet 128: 130–136
9. Padmanabhan V, Cooper K (2000) Concomitant adenoma and hybrid carcinoma of salivary gland type arising in Bartholin's gland. Int J Gynecol Pathol 19: 377–380
10. Seifert G, Donath K (1996). Hybrid tumors of salivary glands. Definition and classification of five rare cases. Oral Oncol Eur J Cancer 32: 251–259
11. Shikhani HA, Shikhani TL, Kuhajda PF, Allam KC (1993) Warthin's tumor-associated neoplasms: a report of two cases and review of the literature. ENT J 72: 264–273
12. Simpson RH (1997) Pitfalls in salivary gland pathology: hybrid epithelial-myoepitehelial carcinoma and adenoid cystic carcinoma (abstract 067). Pathol Res 193: 342
13. Skalova A, Simpson RH, Lehtonen H, Leivo I (1997) Assessment of proliferative activity using the MIB1 antibody help to distinguish polymorphous low grade adenocarcinoma from adenoid cystic carcinoma of salivary glands. Pathol Res Pract 193: 695–703
14. Snyder ML, Paulino AFG (1999). Hybrid carcinoma of the salivary gland: salivary duct adenocarcinoma adenoid cystic carcinoma. Histopathology 35: 380–383
15. Zardawi IM (2000) Hybrid carcinoma of the salivary gland. Histopathology 17: 283–284