Adenoid cystic carcinoma of the tongue: Case report and literature review

Eduardo Costa Studart Soares [1], Fernando Porto Carreiro Filho [2], Fábio Wildson Gurgel Costa [3], Ana Carolina Maciel Jácome Vieira [4], Ana Paula Negreiros Nunes Alves [5]

(1) Doctor of Dental Surgery (D.D.S.), Oral & Maxillofacial Surgeon. Director for the Oral & Maxillofacial Surgery Division of the Walter Cantídio University Hospital. Professor of Stomatology and Oral Surgery at the School of Dentistry of the Federal University of Ceará, Fortaleza, Brazil
(2) Medical Doctor (M.D.), Member of the Head and Neck Surgery Division at the Ceará Institute of Cancer, Fortaleza, Brazil
(3) D.D.S., PostGraduate Student at the School of Dentistry of the Federal University of Ceará, Fortaleza, Brazil.
(4) D.D.S.
(5) D.D.S., Pathologist, Professor of Oral Pathology at the School of Dentistry of the Federal University of Ceará, Fortaleza, Brazil

Correspondence:
Dr. Eduardo Costa Studart Soares
Rua professor Otávio Lobo,
540/1002, Fortaleza, Ceará
Brasil, CEP: 60176-050
Caixa Postal: 3229
E-mail: estudart@yahoo.com.br

Received: 09/10/2007
Accepted: 06/07/2008

Indexed in:
-Index Medicus / MEDLINE / PubMed
-EMBASE, Excerpta Medica
-SCOPUS
-Indice Médico Español
-IBECS

Soares ECS, Carreiro Filho FP, Costa FWG, Vieira ACM, Alves APN. Adenoid cystic carcinoma of the tongue: Case report and literature review. Med Oral Patol Oral Cir Bucal. 2008 Aug 1;13(8):E475-8.

http://www.medicinaoral.com/medoralfree01/v13i8/medoralv13i8p475.pdf

Abstract
Adenoid cystic carcinoma is a common malignant neoplasm originated from salivary glands. The clinical and pathological findings typical of this tumor include slow growth, perineural invasion, and potential local recurrence. It accounts for nearly 2% to 4% of head and neck area tumors. In minor salivary glands it usually affects the palate. We described herein the case of a patient who presented a posterior tongue lesion, which was treated with surgery and radiotherapy as additional treatment. We also carried out a brief literature review on the subject.

Key words: *Adenoid cystic carcinoma, tongue, treatment.*

Introduction
The Adenoid cystic carcinoma is a relatively rare epithelial tumor of the salivary glands. It accounts for about 5% to 10% of all salivary gland neoplasms, representing 2% to 4% of malignant occurrences of the head and neck area. Approximately 31% of lesions affect minor salivary glands, particularly the palate, though they can also be observed in the submandibular and parotid glands (1).
Although it presents a widespread age distribution, peak incidence occurs predominantly among women, between the 5th and 6th decades of life (2). Typical clinical findings include slow growth, local recurrence, perineural invasion and distant metastasis (3). We present a case of adenoid cystic carcinoma of the tongue, and a brief literature review on its clinical, pathological and therapeutic aspects.

Case Report
A 64-year-old systemically healthy female patient presented to the Division of Stomatology, School of Dentistry at the Federal University of Ceará (Brazil), complaining of an asymptomatic volume growth of the tongue, perceived just one week before consultation. The patient reported a long-term smoking habit, and an otherwise insignificant past medical history. Extra-oral examination and palpation revealed cervical lymph node chain with normal size and consistency. On intraoral clinical exam, a firm sessile nodule of about 4 cm in diameter with a similar coloration to that of the buccal mucosa was observed (Figure 1). Based on patient´s history and clinical exams, associated with a rapid growth of the observed lesion, diagnosis of a malignant neoplasm was hypothesized. An

incisional biopsy was carried out and histopathological analysis demonstrated a neoplasm formed by microcystic spaces rounded by atypical hyperchromatic epithelial cells invading the adjacent muscular tissue, compatible with a cribriform variant of adenoid cystic carcinoma (Figure 2). Subsequently, the patient was sent to the Head and Neck Surgery Division of the Cancer Institute of Ceará (Brazil), where hemiglossectomy and cervical lymph node emptying at the third level were carried out (Figure 3A). Post-surgical radiotherapy sessions were also performed. The patient is currently under a post-surgical 16-month follow-up, showing good health and absence of clinically detected metastasis or local alterations (Figure 3B).

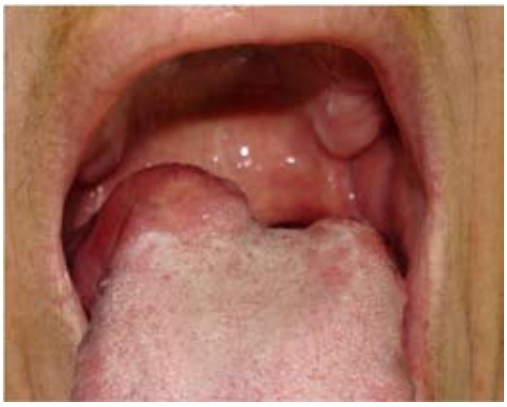

Fig. 1. Nodule elevated in the posterior region of the tongue.

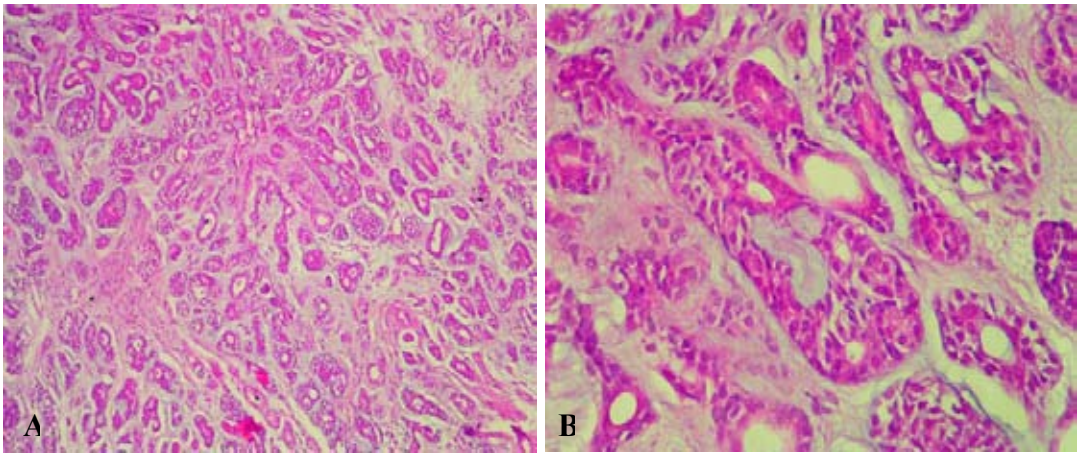

Fig. 2. A) Microphotograph in hematoxylin-eosina stain, 100x zoom, evidencing the formation of microcystic spaces rounded by hiperchromated cells. B) Cribiform characteristic pattern (HE 400x).

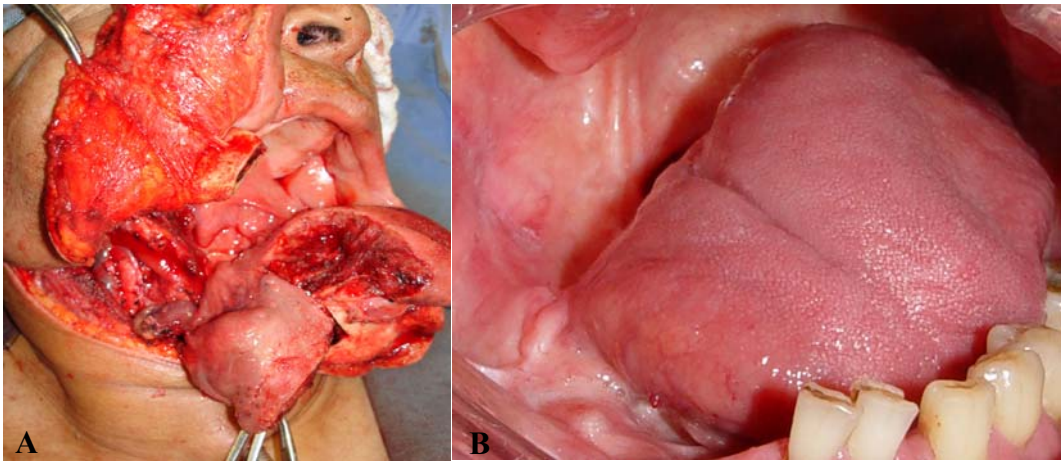

Fig. 3. A) Partial hemiglossectomy after mandibular access and removal of the lesion. B) Post-surgical accompaniment of 16 months after radiotherapy.

Discussion

In 1859, Billroth used the term "cylindroma" to describe the histological standard of four salivary gland tumors. This term was widely used until 1953, when Foote and Frazell renamed the lesion with the expression adenoid cystic carcinoma (4-6). Evesson and Cawson (7) found a discreet predominance of ACC cases in women (F:M 1.2:1) with ages varying from 24 to 78 years. However, most occurrences took place in the seventh decade of life, average age of 66.3 (+13.1). The presently described case agrees with the observations made by these authors regarding age and gender.

The adenoid cystic carcinoma constitutes approximately 29.6% of minor salivary gland tumors. Although the palate is the most commonly involved site (8), nearly 4.4% of cases are located on the tongue (table 1). In a report by Spiro et al. (9) of 242 salivary gland ACC cases, 171 patients presented lesions involving accessory glands, while 64 patients (26%) presented the palate as the affected site, being the tongue the second most affected area. Isacsson and Shear (10) observed neoplasm occurrences in the palate, floor of the mouth, tongue and gingiva, in this order. Goldbatt, Ellis (11) analyzed 55 salivary gland neoplasms of the tongue, and found that 10% of cases were histologically compatible with ACC. De Vries et al. (12) analyzed 178 cases of salivary gland tumors, out of which only 6 cases were located on the tongue.

In these cases differential diagnosis must include benign and malignant tumors related to salivary glands, such as pleomorphic adenoma and adenocarcinoma (8,13).

Treatment of ACC patients has generated great interest and debate among surgeons and pathologists, due to slow clinical development (14). Although it is locally aggressive, with high recurrence levels and late metastasis, commonly leading to patient death between 10 and 20 years after initial treatment (1), compared to other malignant neoplasms, ACC is more difficult to fully remove, with frequently identified positive surgical margins (2).

Possible treatments of ACC include four different modalities (14-17): surgical therapy, radiotherapy, chemotherapy and combined therapy (surgery and radiotherapy, radiotherapy and chemotherapy), being the latter in most cases, the treatment of choice. Only surgical removal or radiotherapy in isolation may fail to eliminate the possibility of recidivation in surgical margins, as well as the occurrence of metastasis in cervical lymph nodes, lungs, bones and brain. (8). In addition, ACC presents a strong neurotropism, with a tendency to invade nerves adjacent to the lesion. In the currently reported case, we have performed an associated therapy (surgical resection and radiotherapy), based on Huang et al. (3) who refers to the potential distant metastasis risk in lesions primarily located in the base of the tongue.

Histologically, ACC can present three different variables: glandular (cribriform), tubular and solid. Mitotic figures are generally scarce in cribriform and tubular areas; however they are easily visualized in solid standards that have been associated with the worst prognosis (17). The relationship between histological pattern and prognosis of adenoid cystic carcinoma has been studied (16,17). Huang et al. (3) observed a survival rate of 16.7 % after a 10 year treatment for cases where solid variable was observed and 47.4% for lesions where cribriform and tubular standards were found. The authors concluded that the main factors associated with patient survival were tumor location, clinical stage and the observed histological variable. Conversely, Spiro et al. (9) have not found histological classification to be of any benefit, and deny a correlation between microscopic appearance and prognosis. Although our ACC patient with cribriform histological standard is clinically stable after 16 months of follow-up, such findings cannot be considered a definitive cure, due to the common occurrence of late metastasis (18). Most studies reveal that ACC presents a bad prognosis and it is therefore necessary to carry out long-term follow-up (19).

Table 1. Series of cases of adenoid cystic carcinoma in minor salivary gland tumors.

Author	Year	Minor salivary gland tumours		Adenoid Cystic Carcinoma	
		n	ACC (%)	Tongue	%
Fine *et al.* (20)	1960	79	13 (16.5)	2	2.5
Leafstedt *et al.*(6)	1971	56	56 (100)	11	19.6
Spiro *et al.* (9)	1974	171	171 (100)	26	15.2
Main *et al.* (21)	1976	112	28 (25)	3	2.7
Isacsson, Shear (10)	1983	201	21 (10.5)	2	1
Eveson, Cawson (7)	1985	336	44 (13.1)	3	0.9
Huang *et al.* (3)	1997	48	48 (100)	10	20.8
Jaber (22)	2006	75	13 (17.3)	3	4
Wang *et al.* (23)	2007	737	143 (19.4)	20	2.7

ACC: adenoid cystic carcinoma

Conclusion

The primary treatment objective in adenoid cystic carcinoma patients is local control, normal functionality and distant metastasis prevention. For this purpose, early detection by the surgeon is a requirement, in order to enable a more favorable prognosis and better quality of life. The therapy involving associated radiotherapy remains the modality of choice for most cases.

References

1. Triantafillidou K, Dimitrakopoulos J, Iordanidis F, Koufogiannis D. Management of adenoid cystic carcinoma of minor salivary glands. J Oral Maxillofac Surg. 2006 Jul;64(7):1114-20.
2. Waldron CA, El-Mofty SK, Gnepp DR. Tumors of the intraoral minor salivary glands: a demographic and histologic study of 426 cases. Oral Surg Oral Med Oral Pathol. 1988 Sep;66(3):323-33.
3. Huang M, Ma D, Sun K, Yu G, Guo C, Gao F. Factors influencing survival rate in adenoid cystic carcinoma of the salivary glands. Int J Oral Maxillofac Surg. 1997 Dec;26(6):435-9.
4. Al-Sukhun J, Lindqvist C, Hietanen J, Leivo I, Penttilä H. Central adenoid cystic carcinoma of the mandible: case report and literature review of 16 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006 Mar;101(3):304-8.
5. Foote FW Jr, Frazell EL. Tumors of the major salivary glands. Cancer. 1953 Nov;6(6):1065-133.
6. Leafstedt SW, Gaeta JF, Sako K, Marchetta FC, Shedd DP. Adenoid cystic carcinoma of major and minor salivary glands. Am J Surg. 1971 Dec;122(6):756-62.
7. Eveson JW, Cawson RA. Tumours of the minor (oropharyngeal) salivary glands: a demographic study of 336 cases. J Oral Pathol. 1985 Jul;14(6):500-9.
8. Maciel Santos MES, Ibrahim D, Neto JC, Da Silva JC, Da Silva UH, Sobral APV. Carcinoma adenóide cístico: relato de caso. Rev Cir Traumatol Buco-Maxilo-Fac 2005; 5:49-54.
9. Spiro RH, Huvos AG, Strong EW. Adenoid cystic carcinoma of salivary origin. A clinicopathologic study of 242 cases. Am J Surg. 1974 Oct;128(4):512-20.
10. Isacsson G, Shear M. Intraoral salivary gland tumors: a retrospective study of 201 cases. J Oral Pathol. 1983 Feb;12(1):57-62.
11. Goldblatt LI, Ellis GL. Salivary gland tumors of the tongue. Analysis of 55 new cases and review of the literature. Cancer. 1987 Jul 1;60(1):74-81.
12. De Vries EJ, Johnson JT, Myers EN, Barnes EL Jr, Mandell-Brown M. Base of tongue salivary gland tumors. Head Neck Surg. 1987 Jul-Aug;9(6):329-31.
13. Ustündağ E, Iseri M, Aydin O, Dal H, Almaç A, Paksoy N. Adenoid cystic carcinoma of the tongue. J Laryngol Otol. 2000 Jun;114(6):477-80.
14. Namazie A, Alavi S, Abemayor E, Calcaterra TC, Blackwell KE. Adenoid cystic carcinoma of the base of the tongue. Ann Otol Rhinol Laryngol. 2001 Mar;110(3):248-53.
15. Moles MA, Avila IR, Archilla AR. Dedifferentiation occurring in adenoid cystic carcinoma of the tongue. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999 Aug;88(2):177-80.
16. Tincani AJ, Del Negro A, Araújo PP, Akashi HK, Martins AS, Altemani AM, et al. Management of salivary gland adenoid cystic carcinoma: institutional experience of a case series. Sao Paulo Med J. 2006 Jan 5;124(1):26-30.
17. Carrasco Ortiz D, Aldape Barrios B. Adenoid cystic carcinoma of the dorsum of the tongue: presentation of a case. Med Oral Patol Oral Cir Bucal. 2006 Aug 1;11(5):E417-20.
18. Pino Rivero V, Pantoja Hernández CG, González Palomino A, Pardo Romero G, Trinidad Ramos G, Blasco Huelva A. Adenoid cystic carcinoma of tongue. Report of a case and literature review. An Otorrinolaringol Ibero Am. 2006;33(5):449-54.
19. Whear NM, Addy JM. Adenoid cystic carcinoma of the sublingual gland-an unusual presentation. Br J Oral Maxillofac Surg. 1993 Apr;31(2):113-6.
20. Fine G, Marshall RB, Horn RC Jr. Tumors of the minor salivary glands. Cancer. 1960 Jul-Aug;13:653-69.
21. Main JH, Orr JA, McGurk FM, McComb RJ, Mock D. Salivary gland tumors: review of 643 cases. J Oral Pathol. 1976 Mar;5(2):88-102.
22. Jaber MA. Intraoral minor salivary gland tumors: a review of 75 cases in a Libyan population. Int J Oral Maxillofac Surg. 2006 Feb;35(2):150-4.
23. Wang D, Li Y, He H, Liu L, Wu L, He Z. Intraoral minor salivary gland tumors in a Chinese population: a retrospective study on 737 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007 Jul;104(1):94-100.