

Association between clinical features, treatment, and recurrence rate of adenoid cystic carcinoma of the palate: a 10-year retrospective study

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Abstract

Adenoid cystic carcinoma (ACC) is both invasive and clinically unpredictable, and our aim was to evaluate the factors that affect its recurrence in the palate. We retrospectively studied 38 patients who had ACC of the palate treated surgically, and the outcome measure was recurrence during the mean (SD) followup time of 55 (18) months. Age, sex, T-stage, bony involvement, duration of follow up, histological type, perineural invasion, and surgical margins were all recorded. Results showed no association between recurrence, and age or histopathological types. However, T-stage ($p=0.001$), sex ($p=0.04$), and bony and perineural involvement ($p=0.01$ in each case) were significantly associated with recurrent tumour. Close superior and posterior margins (<2 mm) were also associated with recurrence ($p=0.001$ in each case). Large tumours with bony and perineural involvement, together with close superior and posterior surgical margins, had a higher risk of recurrence. © 2016 The British Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

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Introduction

Adenoid cystic carcinoma (ACC) is a tumour of the mucus-secreting glandular cells that accounts for about 7.5% of all salivary gland malignancies,^{1,2} and carcinoma of the hard palate makes up about 5% of all cancers of the oral cavity. About 8% of all salivary gland tumours develop in the hard palate, and 80% of them are malignant. Half of all intraoral ACC grow in the palate,³ the incidence of ACC being considerably higher in the palate than in the submandibular and parotid glands.⁴

Histologically there are three types of ACC: cribriform, tubular, and solid. Cribriform is the most common, and solid the least common. ACC are not often found in “pure” types, and it is quite common to have more than one histopathological pattern in a single neoplasm with all three patterns seen in most tumours.³ ACC is known for its prolonged clinical course and tendency to delayed onset of distant metastases.⁵

Clinically the tumour presents as a small, slowly-growing, locally-invasive lesion that overlies extensive subclinical invasion.³ In the early stages metastases are rare. However, distant metastases are important in survival.⁶

Resection is the main treatment for palatal ACC, sometimes together with other treatments, and it is both invasive and clinically unpredictable.⁷ There have been a few studies on the risk factors of recurrent ACC in the palate,⁸ which

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have highlighted some important points - for example, in which part of the tumour do close surgical margins increase the risk of recurrence, and whether or not bony involvement is a risk factor.

The aim of this study was to evaluate the factors that affect the recurrence of ACC of the palate.

Patients and Methods

We designed a retrospective study of patients who presented to the Oral and Maxillofacial Surgical Department, Shiraz University of Medical Sciences between 1 September 2003 and 31 October 2013. The study was approved by the medical ethics committee of the University.

Patients were included in the study if they had histopathologically confirmed ACC of the palate that was treated by resection. They were excluded if they had had a previous operation for ACC or if the palate was involved secondarily from another primary site. Patients were allocated to two groups according to whether the tumour recurred (histologically confirmed), or not, during the follow-up period.

The size of the tumour (T1= <2 cm, T2= 2 cm- 4 cm, and T3= >4 cm), bony involvement (erosion or penetration based on the computed tomographic (CT) scan or magnetic resonance image (MRI) and confirmed during operation), the surgical margin after resection (close margin <2 mm, 2 < 5 mm, or free margin > 5 mm) in five directions (superiorly, posteriorly, anteriorly, medially, and laterally), histopathological type of ACC (cribriform, tubular, or solid) and histopathological involvement of nerves, were the predictive factors of the study. Age and sex were the variables, and local recurrence and distant metastases were the outcomes.

Histopathological type was classified as that seen in more than 70% of the cells during histopathological examination. Surgical margins were reported by oral and maxillofacial pathologists after resection. Patients whose margins were close did not have a second operation. At that time None of the patients was given chemotherapy or radiotherapy during the follow-up period.

Statistical analysis

The statistical analyses were made with the help of IBM SPSS Statistics for Windows (version 19, IBM Corp, Armonk, NY, USA). The chi square test was used to assess the significance of differences between variables in the two groups (recurrence and no recurrence), and the independent *t* test to assess the significance of differences between ages.

Results

We studied 38 patients (Table 1). None of patients initially had lymph node involvement in the neck or distant metastases. Fourteen developed a recurrence during the mean (SD) follow-up period of 55 (18) months (Table 2). All

Table 1

Details of patients (n=38). Data are number (%) except where otherwise stated.

Variable	Measurement
Mean (SD) age (years)	49 (7)
Sex:	
Male	21 (55)
Female	17 (45)
Mean (SD) duration of follow up (months)	55 (18)
Stage of tumour:	
T1	22 (58)
T2	6 (16)
T3	10 (26)
Histological type:	
Cribriform	10 (26)
Tubular	4 (10)
Solid	24 (63)
Bony involvement:	
Yes	29 (76)
No	9 (24)
Perineural involvement:	
Yes	14 (37)
No	24 (63)

recurrences were local. The incidence of recurrences depending on the width of the margins differed significantly posteriorly, medially, and superiorly (Table 3).

Discussion

The palate is the most often involved site for ACC of the minor salivary glands.⁸ It is a slow- growing, aggressive, and destructive tumour⁹ that has a tendency to perineural infiltration, and about 40%-60% of patients have haematogenous spread to distant sites.⁸ It spreads to regional nodes only infrequently.^{10,11} Resection of the tumour with 1-2 cm safety margin is suggested, particularly in the posterior maxilla.^{8,12} The 5-year survival has been reported to be 60%–90%, but 10-year survival is 40%–50%.^{10,13} Variable factors such as size and site of tumour affect recurrence, and reduce survival.^{6,14}

We found no difference in recurrence in various histopathological types, though several studies have reported a better prognosis for the tubular and cribriform forms than for the solid type.^{15,16} More patients who had bone perforated perioperatively developed recurrences than who did not. It seems that bony involvement and perforation are important factors for local recurrence.¹⁷ The size of the tumour has an important influence on recurrence, and it is thought that the most important factors for long-term survival after treatment of ACC are the size and stage of the primary lesion.^{8,18,19} Tumours larger than 4 cm have a higher risk of distant metastases.²⁰ The site of the tumour also complicates treatment and recurrence of ACC. For example, tumours of the maxillary antrum and posterior palate have a poor prognosis because of the complex regional anatomy.⁸ They often present at a locally advanced stage with extensive invasion of nerves and adjacent structures.²¹

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