



REVIEW ARTICLE

## Clinicopathological characteristics and perineural invasion in adenoid cystic carcinoma: a systematic review ☆,☆☆



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### KEYWORDS

Salivary gland neoplasms;  
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### Abstract

**Introduction:** Adenoid cystic carcinoma is the most frequent malignant tumor of the sub-mandibular gland and the minor salivary glands. It is a malignant neoplasm that, despite its slow growth, shows an unfavorable prognosis.

**Objectives:** The aim of this study was to perform a systematic review of the literature on Adenoid cystic carcinoma in the head and neck region and its clinicopathological characteristics, with emphasis on the perineural invasion capacity of the tumor.

**Methods:** A systematic search of articles published between January 2000 and January 2014 was performed in the PubMed/MEDLINE, SciELO, Science Direct, and Scopus databases.

**Results:** Nine articles were selected for this systematic review. These demonstrated that the female gender was more often affected and that malignant tumors showed a high rate of distant metastasis, recurrence, and a low survival rate. The presence of perineural invasion ranged from 29.4% to 62.5% and was associated with local tumor recurrence.

**Conclusion:** Adenoid cystic carcinoma is commonly characterized by the presence of pain, high rate of recurrence, metastasis, and a low survival rate. Reporting studies with patient follow-up is of utmost importance for a better clinical-pathological understanding and to improve the prognosis of this pathology.

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**PALAVRAS-CHAVE**

Neoplasias das  
glândulas salivares;  
Glândulas salivares;  
Neoplasias bucais

**Características clinicopatológicas e invasão perineural do carcinoma adenoide cístico****Resumo**

**Introdução:** O carcinoma adenoide cístico (CAC) é o tumor maligno mais frequente da glândula Submandibular e das glândulas salivares menores. Sendo uma neoplasia maligna, apesar de ter crescimento lento, apresenta um prognóstico desfavorável.

**Objetivos:** O objetivo deste trabalho foi realizar uma revisão sistemática de literatura sobre o carcinoma adenoide cístico na região de cabeça e pescoço e suas características clínico-patológicas com ênfase na capacidade de infiltração perineural do tumor.

**Método:** Uma busca sistemática de artigos publicados entre janeiro de 2000 a janeiro de 2014 foi executada nas bases de dados PubMed/MEDLINE, SciELO, Science Direct e Scopus.

**Resultados:** Nove artigos foram selecionados para realização da revisão sistemática. Nestes, o sexo feminino foi o mais afetado e o tumor maligno apresentou uma alta taxa de metástase a distância, recidiva e baixa taxa de sobrevida. A presença de invasão perineural variou entre 29,4% a 62,5% e foi relacionada à recidiva local do tumor.

**Conclusão:** O CAC é comumente caracterizado pela presença de dor, alta taxa de recidiva, metástase e baixa sobrevida. A realização de estudos com acompanhamento dos pacientes é de extrema importância para uma melhor avaliação clínico-patológica visando melhorar o prognóstico da doença.

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**Introduction**

Salivary gland neoplasms are unusual and account for only about 2–6.5% of tumors of the head and neck region.<sup>1</sup> The frequency of different types of malignant tumors varies according to the site of origin. However, adenoid cystic carcinoma (ACC) appears to be the most common malignant tumor of the submandibular salivary and minor salivary glands.<sup>2</sup>

The ACC is a malignant neoplasm that, in spite of its slow growth, has a poor prognosis due to aggressive tumor invasion and a high rate of recurrence.<sup>3</sup> This neoplasm was first described by Billroth in 1856.<sup>4</sup>

It can develop in several anatomical sites, such as the major and minor salivary glands, lacrimal glands, and upper aerodigestive tract glands.<sup>5</sup> In the buccal-maxillofacial region, it accounts for approximately 22% of neoplasms of major and minor salivary glands, with the minor salivary glands of the hard palate representing the main affected site.<sup>2</sup>

ACC is more prevalent in middle-aged adults and according to many studies, it is more frequent in the female gender.<sup>5,6</sup> Distant metastasis is common; the lung is the most commonly affected site.<sup>7</sup>

It may present clinically as a hardened lump, and pain is an important and common finding in the initial course of the disease. An ulcerated lesion can be seen on the palate, with bone destruction identified radiographically.<sup>8</sup>

Histopathologically, ACC can manifest as different types, with three main recognized patterns<sup>9</sup>: the cribriform and tubular patterns, which are less aggressive, and the solid pattern, which shows cell pleomorphism and mitotic activity, as well as necrotic foci in the central region of neoplastic cell islands.<sup>5,6</sup>

Perineural invasion is a common histological finding, and is considered a possible route for tumor cell dissemination.<sup>5</sup> Perineural involvement occurs in approximately 22–46% of cases of ACC, whether at macro- or microscopic level.<sup>5</sup>

Surgery is the treatment of choice for ACC and may be followed by radiation therapy and, in rare cases, chemotherapy. The frequency of local recurrence for ACC is high, requiring additional surgical resections. Several studies have evaluated this development as a negative one, with the tumor causing the patient's death.<sup>9</sup>

Dental surgeons need to detect any changes in the oral mucosa of their patients. Early diagnosis of ACC results in better quality of life and higher survival rate. The objective of this study was to perform a systematic review of literature on ACC of the head and neck region and its clinical and pathological features, with emphasis on tumor perineural invasion capacity.

**Methods**

A systematic search for articles published between January 2000 and January 2014 was conducted in the PubMed/MEDLINE, SciELO, Science Direct, and Scopus databases. Studies that assessed ACC of the head and neck region and its perineural invasion capacity were evaluated.

The following terms were used in the search: adenoid cystic carcinoma; neoplasm; salivary gland, and perineural invasion, as well as their synonyms and corresponding terms in Portuguese and Spanish, in several combinations. Boolean operators AND, OR, NOT were used when possible. After obtaining the summaries, three independent evaluators selected the relevant studies according to the inclusion and exclusion criteria.

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