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Intra-mandibular adenoid cystic carcinoma

Carcinome adénoïde kystique intra-mandibulaire

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Summary

Intra-mandibular localization of adenoid cystic carcinoma is rare. This tumor is characterized by progressive local, regional, and distant aggressiveness. We reviewed the latest data on this rare type of cancer with a small number of reported cases, a lack of consensus for its treatment, and its bad prognosis.

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Keywords: Mandible, Adenoid cystic carcinoma

Résumé

Le carcinome adénoïde kystique est une tumeur maligne dont la localisation intra-mandibulaire est rare. Il se caractérise par une agressivité progressive locorégionale et à distance. Nous faisons une mise au point sur ce type de cancer rare, avec un nombre réduit de cas, l'absence de consensus pour son traitement et son mauvais pronostic.

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Mots clés : Mandibule, Carcinome adénoïde kystique

Introduction

Adenoid cystic carcinoma (ACC) is a rare malignant tumor of the salivary glands accounting for less than 1% of maxillofacial cancers [1]. The intra-osseous maxillo-mandibular location is very rare [2]. It often develops insidiously and its diagnosis is made at an advanced stage when management can be problematic [1,2]. Moreover, the risk of distant metastases and its neurotropic nature worsen the prognosis [2]. There is no consensus on the therapeutic management due to the rarity of cases. We had for aim to review the latest data regarding intra-mandibular adenoid cystic carcinoma (IACC).

Epidemiology

Twenty-three cases of IACC were reported in the English-language and French-language literature since 1955 (table 1)

[1–10]. IACC generally occurs in the fifth decade of life (table 1) at a mean age of 55.6 years (33–82 years) with a slight male predominance (13 men *versus* 10 women) [1,4–10].

Predisposing factors

Alcohol consumption, smoking, and poor dental health are not predisposing factors for IACC.

Location

IACC location is very rare and the tumor is usually located in the posterior region of the mandibular body and in the angle (22 of 23 cases) [4–9], but the parasymphiseal and symphiseal regions may also be involved (fig. 1) [5,10].

Several hypotheses have been made on the mechanism underlying the atypical intra-osseous location [7]:

- it could be due to either ectopic salivary glands or embryonic remnants of submandibular or sublingual glands trapped within the bone;

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Table I
Literature review on mandibular adenoid cystic carcinoma.

Authors [references]	Sex	Age (years)	Clinical data	Location in the mandible	Treatments	Follow-up
1 - Santos et al. [4]	M	48	Pain – swelling	Molar region	Mandibulectomy and radiotherapy	Unknown
2 - Grimm et al. [5]	W	45	Pain – paresthesia – swelling	Bilateral body and symphyseal	Mandibulectomy Cervical curettage and radiotherapy	Alive at 3 years with metastases
3 - Shamin et al. [6]	M	45	Swelling and metastases at diagnosis	Angle and body	Chemotherapy	Died (unknown delay)
4 - Chen et al. [7]	M	56	Pain	Molar region	Refused treatment	Unknown
5 - Al Shukhun et al. [8]	W	80	Pain	Premolar	Mandibulectomy	Alive at 3 years without recurrence
6 - Gumgum et al. [9]	W	61	Swelling – cervical lymph-nodes	Retromolar	Mandibulectomy cervical curettage and radiotherapy	Unknown
7 - Clark et al. [10]	M	54	Swelling – cervical lymph-nodes	Parasymphyseal	Mandibulectomy cervical curettage and radiotherapy	Alive at 4.5 years without recurrence
8 - Bumstead [11]	M	54	Pain – swelling	Body	Surgery Radiotherapy	Lung metastasis (unknown delay)
9 - Bradley [12]	M	64	Trismus – pain – swelling	Angle	Surgery	Unknown
10 - Hamori et al. [13]	M	43	Otalgia	Body	Surgery	Lung metastasis (unknown delay)
11 - Shin et al. [14]	W	59	Trismus – pain	Angle	Unknown	Unknown
12 - Dahwan et al. [15]	W	40	Swelling	Ramus	Unknown	Lung metastasis (unknown delay)
13 - Slavin et al. [16]	M	35	Swelling	Angle	Surgery	Unknown
14 - Burkes et al. [17]	W	50	Pain	Body	Surgery	Lung metastasis (unknown delay)
15 - Yoshimura et al. [18]	W	47	Paresthesia – loose teeth	Body	Radiotherapy	Unknown
16 - Mushimoto et al. [19]	M	24	Swelling	Body	Surgery	Unknown
17 - Kaneda et al. [20]	M	47	Swelling Loose teeth	Body	Surgery	Unknown
18 - Gingell et al. [21]	W	72	Pain	Body	Surgery	Alive at 7 years without recurrence
19 - Hirota et Osaki [22]	M	82	Swelling	Body	Radio-chemotherapy	Died (unknown delay)
20 - Brookstone et al. [23]	W	33	Pain – trismus	Body angle	Surgery and radiotherapy	Unknown
21 - Favia et al. [24]	W	46	Pain – swelling	Body	Surgery	Alive at 14 years without recurrence
22 - Chen et al. [25]	M	56	Pain	Body	Surgery	Unknown
23 - Our case	M	57	Pain – swelling – loose teeth	Symphyseal Body	Mandibulectomy, cervical curettage and radiotherapy	Alive at 6 months without recurrence

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