

原文題目(出處)：	New classification of maxillary ameloblastic carcinoma based on an evidence-based literature review over the last 60 years Head & Neck Oncology 2009;1:31
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內文：

### Abstract

- **Background:**
  - ameloblastic carcinoma rather occurs in the mandible than in the maxilla.
  - Its rarity and speculative histopathogenesis may account for diagnostic difficulties.
  - Current classifications do not consider
    - ◆ benign histopathological features at the primary
    - ◆ malignant features at the metastatic tumour site.
  - Based on an evidence-based literature review, a recommendation for a novel classification is presented.
- **Methods :**
  - An evidence-based literature review over the last 60 years regarding ameloblastic carcinoma of the maxilla was conducted.
- **Result:**
  - 26 cases (mean age: 54.4 (583 years); male to female ratio: 2.7to 1).
  - 54% : the primary diagnosis was ameloblastic carcinoma
  - 34.6% : revealed pulmonary metastases
  - only one : cervical lymph node metastasis
  - two : no malignant histopathology at the primary but at their metastatic sites.
  - 73,1% : controlled during a median followup time of 54,3 months  
(6 to 156 months)
  - 6 : died of disease after a median time of 62,7months after initial diagnosis.  
(7 to 156 months)
- **Conclusion :**
  - be aware of : ameloblastomas may degenerate into a "malignant" disease with recurrence and metastasis.
  - special attention should be paid to potential pulmonary involvement.

### Introduction

- **Ameloblastomas:**
  - 1% of all jaw tumours,
  - benign, but locally aggressive odontogenic epithelial neoplasms
  - differential diagnoses:
    - ◆ calcifying epithelial odontogenic tumor
    - ◆ metastatic carcinoma of the jaw
    - ◆ keratocystic odontogenous tumours
  - maxillary : mandibular = 1 : 5
  - most common site : mandibular molar region
  - > 50% recurrence rate within the first 5 years after primary surgery
  - > 3600 cases of ameloblastomas ; < 60 cases of ameloblastic carcinoma  
( mandible 2/3)

→ little is known about their malignant features
- **Ameloblastic carcinoma:**
  - malignant ameloblastoma V.S. ameloblastic carcinoma.
    - ◆ ameloblastic carcinoma: malignant histopathological features  
independent of the presence of metastasis
    - ◆ malignant ameloblastom : metastasize as well differentiated benign cells
  - 70~85% metastases of ameloblastoma occur in the lungs
- **Maxillary ameloblastic carcinomas have not been well studied**
  - published data of the maxilla : single-case reports
  - lacking of prospective data or data from multi-centre studies
  - lack of results from long-term follow-up
    - ◆ most case reports cover less than 5 years after the primary operation.
    - ◆ long-term follow-up is not possible due to advanced age of patients.
- **Aim :**
  - collect clinical features and treatment results of maxillary ameloblastic carcinoma over a period of 60 years in order to implement a novel classification for this type of carcinoma.

### Methods

- Electronic databases (Medline and Cochrane) were searched
  - using a set of predetermined keywords.
  - developed and implemented for PubMed
  - No restriction was placed on the year or language of publication.
- The search strategy was devised in consultation with a senior librarian.
  - The citations retrieved from each database were exported into the

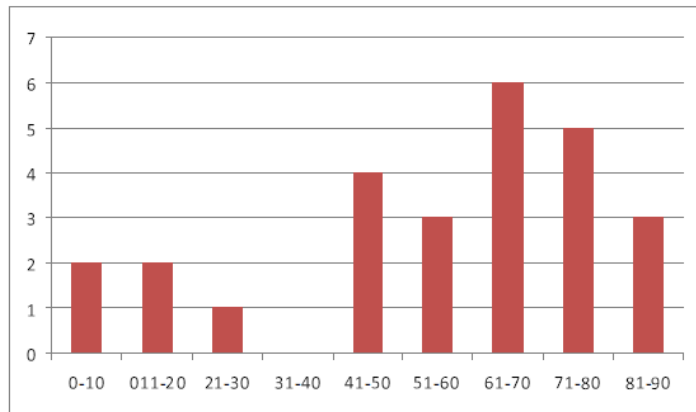
*EndNote bibliometric management software.*

- The titles and abstracts were screened
- hard copies of all potentially relevant articles were retrieved.
- Their reference lists were manually searched for any related articles.

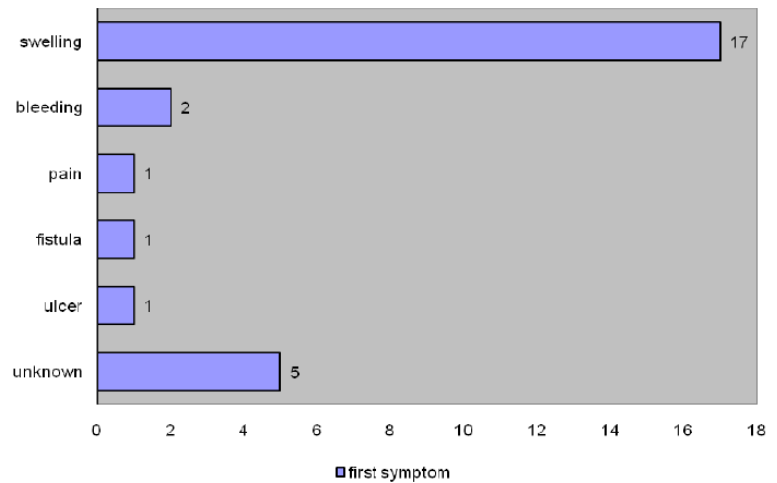
**Result**

● 26 cases of maxillary ameloblastic carcinomas

- mean age : 54.4 years
- marked prevalence : 41 ~ 80 years (69.2%)
- female : male = 1 : 2.7



- chief complaint : swelling

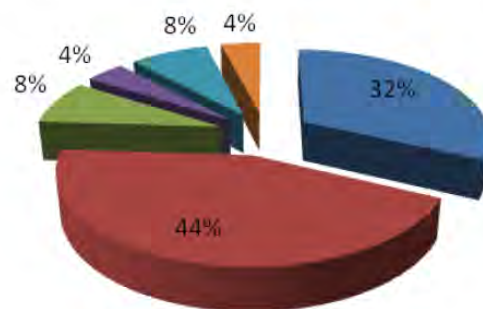


- tumour localization :

44% left maxilla,  
32% right maxilla

**primary localization**

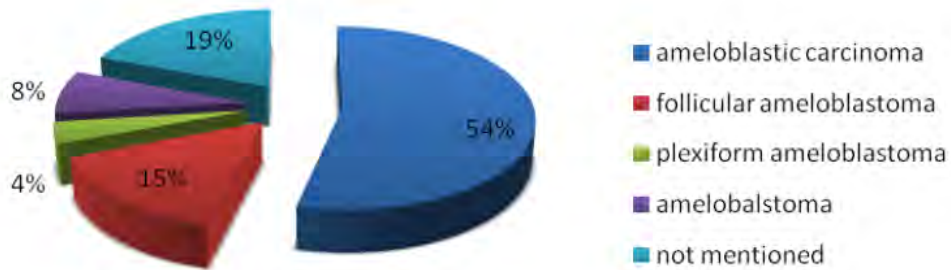
- right maxilla
- left maxilla
- anterior maxilla
- left maxillary sinus
- right maxillary sinus
- no particular location



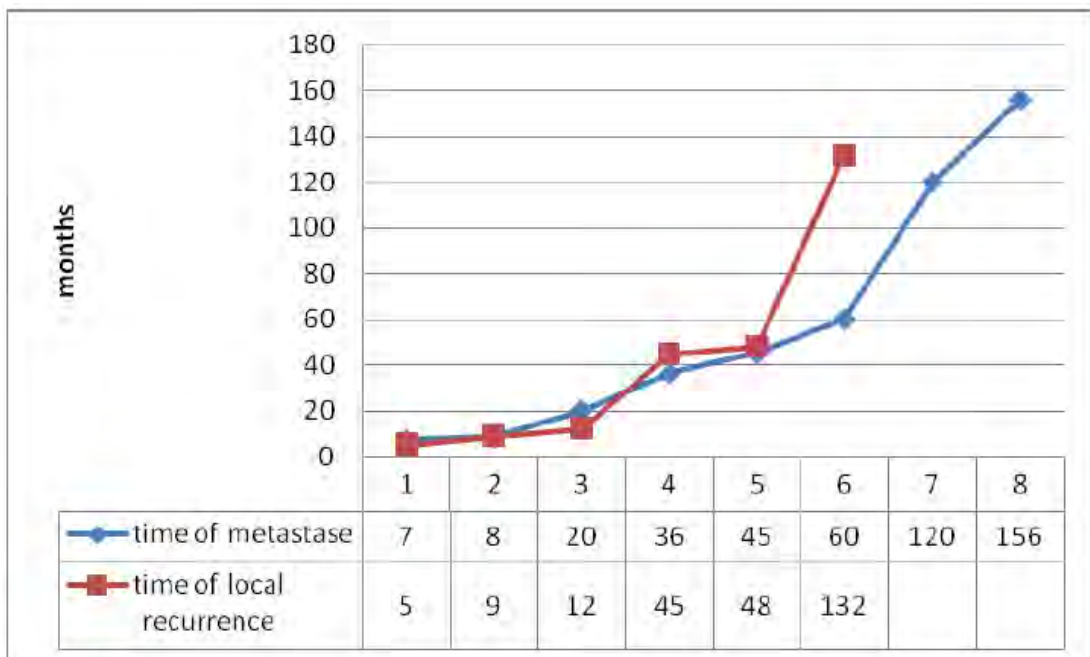
- first specimen : 54%

ameloblastic carcinoma, 15% follicular ameloblastoma

### primary diagnosis



- pulmonary metastases : 26,9%
- local recurrence : 23%
- 77% underwent follow-up period of 54.3 months (6~156 months)
- 23% died of disease after 62,7 months (7 months-10 years)



### Discussion

- **intention of this study :**
  - collect aspects of clinical appearance
  - compare these results with the current classifications of ameloblastic carcinomas.
  - The first clinical sign
    - ◆ 61.5% : swelling; bleeding
    - ◆ 15.4% : ulceration or fistula

- Progressive types of ameloblastic carcinomas
  - degree of aggressiveness
    - ◆ cortical bone perforation
    - ◆ invasion of soft tissue
    - ◆ recurrences
    - ◆ metastases
      - Both pathways : haematogenous / lymphatic(rare)
  - Among these cases:
    - ◆ metastases : 34.6%
    - ◆ local recurrences : 23.1%
    - ◆ neck lymph nodes involved : only one
    - ◆ pulmonary metastases : 26.9%
  - detect pulmonary metastases
    - ◆ computertomography
    - ◆ PET-scans
    - ◆ long-term follow-up
    - ◆ increasing serum calcium (predictor of metastases)
  - two factors predictors for metastasis
    - ◆ granular cell change;
    - ◆ extensive clear cell
    - ◆ these histopathological features have not been investigated
    - ◆ general interpretation is lacking evidence
    - ◆ difficulty: whether the carcinoma is a
      - different entity
      - originated from an ameloblastoma
- In the present study
  - 50% was determined to be an ameloblastic carcinoma
  - not excluding the potential development of an undetected ameloblastoma
  - Especially this problem has not been considered in current classifications
- A novel classification
  - A significant disadvantage : the origin including the histopathogenesis of ameloblastic carcinoma is still unknown
  - modification of the current classification
    - ◆ primary ameloblastoma is followed by secondary metastasis with histopathological features of malignancy without evidence of malignancy in the primary localization

Table 2: Classifications of ameloblastic carcinoma by Elzay (1982) and Slootweg & Müller (1984) and the novel classification

Type	Elzay (1982)[32]	Slootweg & Müller (1984)[33]	Kruse et al.(2009)
1	<b>Arising from an odontogenic cyst</b>	Primary intraosseous carcinoma ex odontogenic cyst	Malignant ameloblastoma
1a	-	-	Metastase with features of an ameloblastoma (well differentiated)
1b	-	-	Metastase with malignant features (poorly differentiated)
2	<b>Arising from an ameloblastoma</b>		Ameloblastic carcinoma arising from an ameloblastoma
2a	Well differentiated (malignant ameloblastoma)	Malignant ameloblastoma	Without metastase
2b	Poorly differentiated (ameloblastic carcinoma)	Ameloblastic carcinoma, arising <i>de novo</i> , ex ameloblastoma or ex odontogenic cyst	Metastase with features of an ameloblastoma (well differentiated)
2c			Metastase with malignant features (poorly differentiated)
3	<b>Arising <i>de novo</i></b>	Primary intraosseous carcinoma <i>de novo</i>	Ameloblastic carcinoma with unknown origin histology
3a	No keratinizing	No keratinizing	Without metastase
3b	Keratinizing	Keratinizing	Metastase with features of an ameloblastoma (well differentiated)
3c			Metastase with malignant features (poorly differentiated)

- **treatment of maxillary ameloblastomas**
  - treated as radically as possible
    - ◆ due to the spongy maxillary bone architecture
      - may facilitate the spread of the tumour
      - may lead to infiltration of adjacent vital structures.
    - ◆ the speed of growth in the mandible is decelerated
      - due to the thick and compact bone structure
  - A surgical resection with 10~15 mm margin free of tumour
  - even though may be limited related to adjacent pivotal anatomical structures
  - Regular follow-up and CT- or MRI controls
- **Controversy regarding its treatment:**
  - Some authors have suggested radiotherapy
  - Others doubt its effectiveness
  - Philip et al. (2005) suggested to apply adjuvant radiotherapy in patients
    - ◆ with positive resection margins
    - ◆ multiple positive lymph nodes
    - ◆ extracapsular spread
    - ◆ perineural invasion
    - ◆ salvage surgery would be inefficient

- Reports about chemotherapy are rare.
  - ◆ only 3 patient and all of them died.
- Among all the reviewed cases
  - 27% presented
    - ◆ at first diagnosis as a follicular or plexiform ameloblastoma
    - ◆ secondarily as an ameloblastic carcinoma

**Conclusion**

- novel classification :
  - considers the unknown origin
    - ◆ primary ameloblastomas with metastases
    - ◆ their histopathological features of malignancy
    - ◆ without previous evidence of malignancy in the primary localization.
- Treatment in cases of maxillary ameloblastomas
  - a radical resection should be performed
    - ◆ prevent recurrence and development of malignancy.
  - Patients should undergo a life-long follow-up
    - ◆ including regular CT or MRI scans,
    - ◆ for early detection of recurrence.
  - For the staging procedure PET scan or chest CT should be performed
    - ◆ detect pulmonary metastases.
  - A neck dissection
    - ◆ only be performed in the presence of clinically positive lymph nodes.

題號	題目
1	Which statement of ameloblastoma was wrong? (A) Are tumors of odontogenic epithelial origin (B) Are slow-growing, local invasive tumor (C) May arise from a developing enamel organ (D) More exist on maxilla than mandible
答案()	出處：Oral & Maxillofacial Pathology P.611
題號	題目
2	Malignant ameloblastoma V.S ameloblastic carcinoma Which was wrong? (A) ameloblastic carcinoma shows features of malignancy (B) Malignant ameloblastoma most often metastases to liver (C) The prognosis of Malignant ameloblastoma is poor (D) x-ray finding of ameloblastic carcinoma shows ill-defined margin and cortical destruction
答案()	出處：Oral & Maxillofacial Pathology P.619