

原文題目(出處)：	An incidentally discovered radiolucency in the posterior mandible. Oral Surg Oral Med Oral Pathol Oral Radiol 2012;113;17-20.
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內文：

## Clinical Presentation

### Personal data

- (1) Gender : female
- (2) Age : 61 y/o

### Chief complaint

→ a radiolucent lesion is found during routine oral examination over left retromolar region

### Past medical history

- (1) type II DM (managed with metformin)
- (2) alcohol (-)
- (3) tobacco (-)
- (4) drug use (-)
- (5) No remarkable family history for neoplastic disease

### Past dental history

- (1) Restoration over several decades
- (2) Extraction of 38 without periapical pathology at 19 y/o

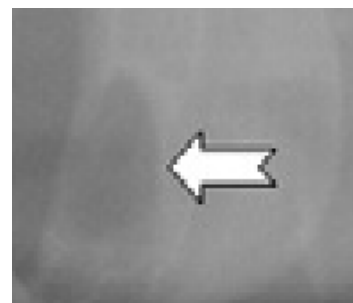
### Clinical examination

- (1) Normocephalic atraumatic head with normal range of mandibular movement
- (2) Occlusion : stable
- (3) No lymphadenopathy is noted over cervical region
- (4) No soft tissue pathology is noted over intraoral area

### Radiography examination

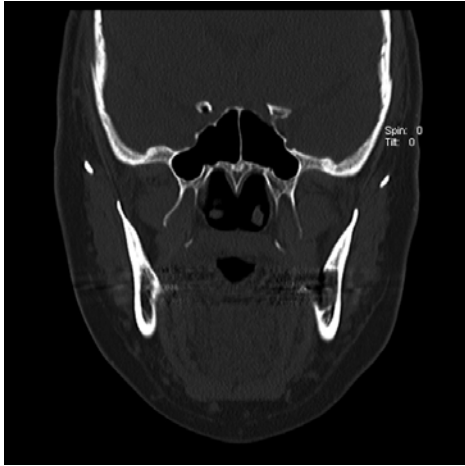
- (1) Panoramic finding

A well circumscribed noncorticated radiolucent lesion distal to 37, within the ramus of left mandible about 1.5 x 2.0 cm in diameter



- (2) CT finding

a osteolytic lesion centered within left ramus , penetrating the lingual plate



## Differential Diagnosis

A significant number of intrabony jaw lesion have their origin from tooth-forming tissue → odontogenic cysts and odontogenic tumors are logical start for differential diagnosis

### Odontogenic Cysts

→ Keratocystic odontogenic tumor

1. affect mandible (75%)
2. strong propensity for posterior region
3. between 10~40 y/o
4. well corticated borders, sometimes(38%) associated with an unerupted tooth or earlier extraction site

→ Residual cysts would not be considered because of the conditions

### Odontogenic Tumors

Well-circumscribed → a benign tumor or a low-grade malignancy

→ Odontogenic myxoma

1. equal predilection for maxilla and mandible (23% in posterior mand.)
2. a “soap-bubble” appearance spanning from the premolar region to the molars  
--- typical feature of myxoma

→ Ameloblastoma

1. common in mandible(80%)
2. affect molar-ramus area(39%~66%)
3. average age : middle to 30s , just 10% in 70s
4. large R/L lesion with bone expansion and “honeycombed” appearance
5. sometimes contains an unerupted tooth (usually third molars)

### Nonodontogenic neoplasm

→ Desmoplastic fibroma

1. 84% found in mandible and 70% lesions in mandible affect ascending ramus
2. 84% occur in people younger than 30 y/o

→ Neurofibroma

1. common on buccal mucosa and dorsum of tongue , sometimes in bone

### Malignancy

→ Metastatic disease

1. usually symptomatic
2. not uncommonly an oral metastasis can precede the discovery of the primary

site

→ Clear cell odontogenic carcinoma

1. uncommon
2. 80% in mandible
3. usually found in people elder than 50 y/o

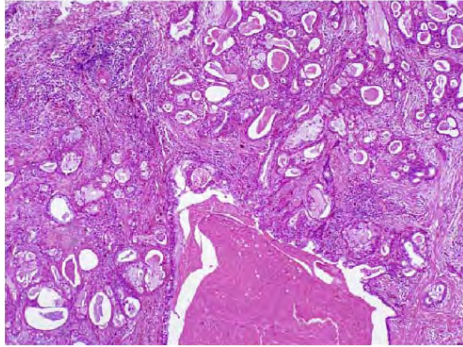


Fig. 3. Photomicrograph showing small infiltrating cords and islands of neoplastic epithelium with micro- and macrocystic areas. Hematoxylin and eosin stain. Original magnification ×13.

4. cortical bone perforation, soft tissue involvement are noted

→ Primary mucoepidermoid carcinoma

1. may associate with ectopic salivary gland, odontogenic epithelium(mucus-producing cells)
2. rarely occur
3. middle-age adults and slightly female predilection
4. common in mandible (often in molar-ramus area)
5. cortical swelling, sometimes bone destruction is noted

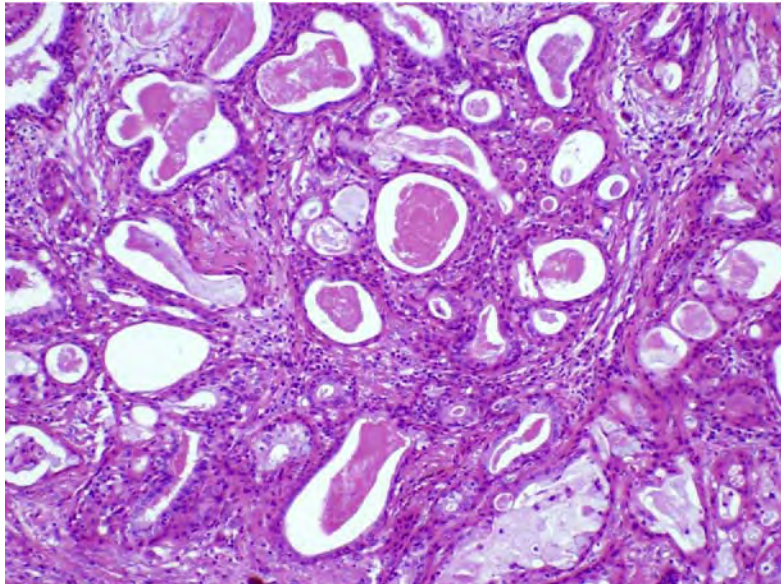
## Diagnosis

(Incisional biopsy is operated)

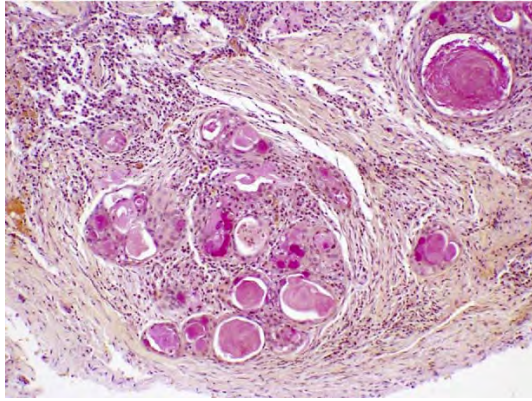
### Histologic examination

(1) numerous nets and large sheets of epithelial cells with both microcystic and macrocystic area

well-formed mucus cells were mixed with the epidermoid cells and mature squamous differentiation was noted. Mitoses were rarely encountered and perineural invasion, necrosis, and high-grade cytologic atypia were absent



(2) mucicarmine special stain demonstrated intracytoplasmic staining of mucous cells



### Positron-Emission tomography finding

- (1) no indication of metastatic disease throughout the body
- (2) no suggestion of another primary neoplasm

Final diagnosis → intraosseous mucoepidermoid carcinoma

## Management

### Surgical treatment

- resected with 1-cm safety margin
- buccal resection : subperiosteal with cortical plate intact
- lingual resection : supraperiosteal, including lingual mucosa and sacrificing lingual nerve
- coronoid process and condylar process remain intact

### Post-OP therapy

- IMF for second-stage surgery and for accurate reconstruction of the mandibular continuity
- harvesting a bicortical bone graft from iliac crest
- reconstruction of the ascending ramus with a 2.3 mm Stryker Leibinger fixation plate

### 12-month follow up

- radiography shows osteogenesis over the donor tissue and the recipient site
- Occlusion : stable
- MMO : 37 mm

## Discussion

Primary intraosseous adenocarcinoma is rare, but when it occurs, most often be confined in jaw, particularly the mandible(ramus and body).

### The 3 most common subtype of intraosseous adenocarcinoma

- (1) MEC (most prevalent)
- (2) Adenoid cystic carcinoma
- (3) Adenocarcinoma not otherwise specified
- fewer than 200 cases have been reported in literature, the majority of Which(n=135) are intraosseous MEC

### The origin of central salivary gland tumors

- (1) developmental remnants of submandibular salivary gland
- (2) ectopic entrapment of retromolar minor mucous glands
- (3) glandular metaplasia of epithelial rest of dental lamina
- (4) expression of glandular potential of the epithelial lining of odontogenic cyst



- in this case, due to the lack of details, it's hard to rule out the origin
- if it's odontogenic origin, 32%~48% of central MEC are associated with an Impacted tooth or an odontogenic cyst

**Genetic analysis**

- soft tissue and MEC with the chromosomal translocation
- CRTC1/MAML2, increasing likelihood of metastasis

**Clinical and Radiography features of central MEC**

- (1) no sexual predilection
- (2) from first to seventh decade of life(predilection of middle age)
- (3) affect 3 times in mandible than in maxilla
- (4) posterior mandible, rarely in anterior jaws
- (5) usually asymptomatic, but if the neoplasm expanding, pain and swelling may occur.
- (6) Unilocular or multilocular RL, well or ill-defined (often well-defined)
- (7) The margins are noncorticated, but typically the cortical plate is intact

**Diagnosis**

- cortical plates are usually intact
- A clinical 3-stage classification for classifying central MECs
- cortical perforation and destruction of bone → stage III

**Treatment**

- (1) Aggressive surgical resection
  - En bloc resection
- (2) Conservative approach
  - enucleation, curettage, marsupialization combined with RT

**Prognosis**

- En bloc resection : recurrence less than 4%
- Conservative approach : recurrence 40%
- Survival rate of 2- and 5-year f/u after aggressive treatment = 100%
- Metastasis from maxilla = 0 / from mandible = 39% to cervical region before Treatment (cytogenetic analysis of soft tissue shows high correlation with CRTC1/MAML2 fusion and metastasis)

**Summary**

- (1) The intraosseous MEC in this case is an asymptomatic, well-circumscribed, noncorticated radiolucency of the retromolar region of left mandible
- (2) Differential diagnosis of the lesion contain primary odontogenic cysts and tumors, and nonodontogenic tumors or metastasis
- (3) The histogenesis of intraosseous MECs is still debated
- (4) Surgical treatment of this case is associated with good prognosis

題號	題目
1	Case中採用En bloc resection切除左側lingual nerve後，對於patient術後哪項是可預期的狀況 (A) P't 左側舌頭從此活動不便，失去部分口腔自淨能力 (B) P't左側舌頭從此對於酸甜鹹的味覺不靈敏 (C) P't左側舌頭從此失去痛覺，可能有ulcer而不自知 (D) 無關緊要，因舌頭是由Glossopharyngeal nerve支配
答案(C)	出處：Contemporary Oral and Maxillofacial Surgery,5 <sup>th</sup> ed.
題號	題目

2	有關MEC治療的預後敘述，以下何者錯誤
	(A) 若顯微鏡下發現cyst formation越少，而degree of cytologic atypia越高，則預後越差 (B) 整體而言，intraosseous MEC的預後佳，P't死亡常因MEC metastasis而非recurrence (C) Solid island of squamous cell and intermediate cells為high-grade MEC的特徵 (D) 治療方式的選擇隨MEC的惡性程度高低而有所不同，所以術後combine RT為非必需的
答案(B)	出處：Oral and Maxillofacial Pathology