

原文題目(出處)：	Mixed choristoma on the anterior dorsal tongue: a new case and review of the literature. Oral Surg 2011;4:26-9
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<內文>

### **Introduction**

- A choristoma is a benign tumour-like growth consisting of normal tissue in an abnormal site, which should be included in the differential diagnosis of any firm swelling on the tongue.
- 1913 Monserrat
  - First reported osseous choristoma on the tongue and used the term 'lingual osteoma'
    1. The lesion behaves benignly and can be treated by surgical excision with extremely rare recurrence.
- 1971, Krolls et al.
  - Introduced a more appropriate term 'osseous choristoma'
- This paper
  - Describe the clinical and microscopic features of osseous and cartilaginous choristomas with brief review of the literature.
  - Report the second case of mixed osseous choristoma to occur on the anterior dorsal tongue.

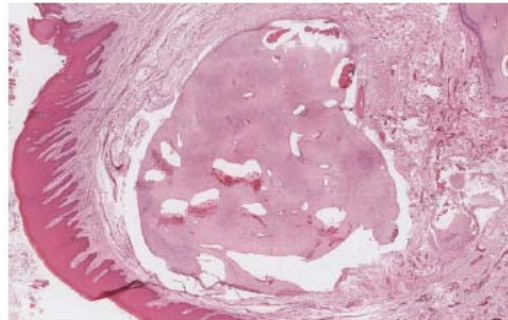
### **Case report**

- First visit : May 2008
- Patient's data : 57-year-old male
- S : asymptomatic pedunculated swelling on the anterior right dorsal aspect of the tongue for 3 years and could not elicit a history of trauma.
- Lesion
  - 0.8 cm in greatest dimension
  - Blanched slightly under pressure but appeared to arise from the underlying connective tissue
  - Surface mucosa : slightly keratotic showing evidence of minor chronic trauma
  - Hard on palpation.

- Clinically
  - Traumatic fibroma (fibroepithelial polyp) although a peripheral ossifying fibroma was also entertained



**Figure 1** Firm soft tissue lesion located on the anterior right dorsal tongue.



**Figure 2** Histological section reveals a polypoid mass of well encapsulated cartilage and dense bone beneath a non-ulcerated non-papillated lingual mucosa (final magnification  $\times 100$ ; stain H&E).

- Treatment
  - Excised completely under local anaesthesia
  - Examined by routine histopathology with haematoxylin and eosin
  - Lesion healed well without complication
- Microscopic examination
  - Polypoid mass of well encapsulated cartilage and dense bone beneath a non-ulcerated non-papillated lingual mucosa
  - No evidence of active chondroblasts, osteoblasts or osteoclasts
- Histology diagnoses
  - Osseous choristoma with cartilaginous components

## Discussion

- Osseous choristomas and cartilaginous choristomas are relatively rare benign lesions with approximately 61 and 28 cases having been reported in the literature, and only 7 cases reported to contain a mixture of these tissues
- **Osseous lesion**
  - Male to female ratio : 1:5
  - Hard pedunculated smooth surfaced lesions
  - Site :
    - ✓ Base of the tongue adjacent to foramen caecum and the circumvallate papillae
    - ✓ No case reported occurring at ventral tongue
    - ✓ Other occurring site : buccal mucosa, submental region, retromolar pad area and masticatory muscles
  - Presentation ranges(lingual lesion) :
    - ✓ Age at time of initial diagnosis ranging from 8 to 73 years
  - Diameter(lingual lesion) :
    - ✓ Generally less than 2.5 cm in diameter

- Histologically
  - ✓ Lesion consists of a well circumscribed, lamellated mass of dense viable bone with a haversian system, as well as osteocytes in lacunae
  - ✓ Bony mass is surrounded by dense fibrous connective tissue covered with thin stratified squamous epithelium
- **Cartilaginous lesions**
  - No difference in male to female ratio
  - Site :
    - ✓ Dorsal tongue, but four cases have been reported on the ventral tongue
    - ✓ Other occurring site : gingiva, soft palate, buccal mucosa and palatine tonsi
  - Presentation ranges :
    - ✓ Between 3 to 75 years of age
    - ✓ Most commonly during the third and fourth decades of life
  - Diameter :
    - ✓ 4.5 cm in greatest
  - Microscopically
    - ✓ Lesion consist of a circumscribed nodule composed of hyaline cartilage with well-defined lacunae, showing small chondrocytes
- **Mixed osseous and cartilaginous components**
  - Male to female ratio of 6:1
  - Site :
    - ✓ Base of the tongue (4 cases, 57%)
    - ✓ Ventral tongue (2 cases, 28%)
    - ✓ Anterior third of the tongue (1 case, 15%)
  - Presentation ranges :
    - ✓ From 20 to 67 years
  - Diameter :
    - ✓ Varies from 0.7 to 2.5 cm

**Table 1** Summary of the main clinical data of mixed lingual choristoma cases

Authors	Sex	Age (years)	Location	Diameter (cm)	Duration	Symptom
Roy <i>et al.</i> <sup>28</sup>	F	20	Near foramen caecum	1.0	10 years	Bothersome
Gabriele & Kaufman <sup>29</sup>	F	22	Near foramen caecum	1.0–1.5	Found during routine examination	Asymptomatic
Wesley & Zielinski <sup>30</sup>	F	57	Ventral surface of the tongue	1.0	Entire life	Asymptomatic
Landini <i>et al.</i> <sup>24</sup>	F	35	Left lateral border of the tongue	0.8	23 years	Asymptomatic
Watson <i>et al.</i> <sup>31</sup>	F	67	Junction between anterior one third and posterior one third	0.7–1.0	6 months	Sensation of a lump in the throat
Piattelli <i>et al.</i> <sup>32</sup>	F	64	Right ventral tongue	0.8	2 years	Not mentioned
Lee <sup>33</sup>	M	26	Posterolateral border of the tongue	2.5	8 years	Asymptomatic
AbdulMajeed & Farah	M	57	Anterior right dorsal aspect of the tongue	0.8	3 years	Asymptomatic

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- Choristomas of the tongue are mostly asymptomatic
  - Some reports of dysphagia, gagging, nausea, irritation and swelling in the throat.
  - The present case : asymptomatic
  - Differential diagnosis :
    - thyroid nodule, hyperplastic tonsils, fibroma, granular cell tumour and neural neoplasm
  - The present case : consistent fibroma (location and tissue consistence)
  - Treatment :
    - surgical excision, with recurrence being extremely rare
    - No follow-up complication of lingual choristomas has been described in the literature
  - Aetiology 病因學 (developmental or reactive)
    - Developmental theory
      - ✓ complex embryology of the tongue leads to a developmental malformation which includes ossification of the branchial arch remnants or calcifying of the thyroid gland remnants
      - ✓ explain why osseous choristomas are so widely distributed in the area of foramen caecum and the circumvallate papillae
      - ✓ cartilaginous choristomas →
        - from heterotopic cartilage remnants from any of the first four branchial arches
        - chondroblastic cells are misplaced during development and sequestered in the tongue
    - reactive theory
      - ✓ lesion is reactive in nature →
        - central ossification similar to myositis ossification
      - ✓ In the case of cartilaginous choristomas, the reactive theory postulates that trauma can stimulate metaplastic change
  - The present case :
    - branchial remnants or thyroid tissue were not found histologically
    - history and the position of the lesion on the anterior third of the dorsal tongue makes trauma the most possible cause
    - suggested :
      1. traumatic event such as a tongue bite that led to chronic inflammation
      2. stimulating metaplastic change to fibrous, cartilaginous and finally to osseous tissue
    - explained :
      - ✓ lesion contained a mixture of cartilaginous elements within a solid
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osseous mass

- Previous literature has considered osseous and cartilaginous choristomas as separate entities
- The paper propose that these two types of choristoma have a common aetiopathogenesis
  - Traumatic episode
    - local inflammation
    - development of hyperplastic tissue, and fibroma formation
    - further trauma
    - metaplastic ossification of a degenerating fibroma begins with cartilaginous formation
    - formation of bone within the lesion
- osseous and cartilaginous choristomas have the **same aetiopathogenesis**
- osseous choristomas represent a **late presentation** of the lesion
- explain why most choristomas reported in the anterior part of tongue are cartilaginous, as they are more likely to be identified by the patient or clinician early, with ensuing diagnosis and surgical removal.
- lesions develop at the base of the tongue : have more time for osseous formation given that they are asymptomatic and not identified at an early stage

**conclusion**

- osseous and cartilaginous choristomas should be included in the differential diagnosis of any firm swelling on the tongue
- lesion may occur on any region of the tongue
- osseous choristomas represent a late stage in the development
- Awareness of its benign behaviour is important to avoid aggressive surgical management.

題號	題目
1	Which disease is not a tumorlike growth of microscopically normal tissue in an abnormal location? (a) Choristoma (b) Soft tissue osteomas (c) Fibroma (d) Soft tissue chondromas
答案(c)	出處 : Oral and maxillofacial pathology (third edition) p552
題號	題目
2	Where is most common location of the choristoma? (a) Posterior tongue near foramen cecum (b) Buccal mucosa

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	(c) Lip (d) Gingiva
答案(a)	出處：Oral and maxillofacial pathology (third edition) p552