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설배부에 발생한 연골성분리종 1례

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A Case of Chondroid Choristoma on the Dorsum of the Tongue

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= Abstract =

Chondroid choristomais a rare tumor like lesion of normal tissue in an unusual location. Oral cavity chondroid choristoma is exceedingly uncommon. This lesion is commonly covered by normal oral mucosa and can develop during a whole lifetime. We experienced a case of 57-year-old man who presented as 6-months history of asymptomatic mass on the dorsal surface of the tongue. We performed surgical excision under local anesthesia, and the pathological diagnosis was chondroid choristoma. After surgery, patient was followed up without any recurrence and discomfort. Therefore, we report this case with a review of literature.

Key Words : cartilage, choristoma, tongue

Introduction

Choristoma is a relatively unusual tumor-like mass composed of normal tissues like dermal and epidermal components, muscle, cartilage, and bone that has arose in an abnormal site.[1)] The most common involved sites of chondroid choristoma are small joints of hands and feet.[2)] This cartilage producing choristoma is rare in the oral cavity mucosa.[3)] Tongue is the most common involved site, although this tumor has also been located in the gingiva, soft palate, and buccal mucosa.

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Chondroid choristoma can develop during a whole lifetime.[1)] It frequently appears as solid, painless mass. The tumor has a slow-growth pattern and rarely exceeds a few centimetres in diameter.[2)] The histopathological appearance of chondroid choristoma is composed of mature hyaline cartilage, bone and fat tissue in various proportions.[4)] The treatment of a chondroid choristoma consists of surgical excision of the tumor.[5)]

Here we present chondroid choristoma in the tongue and discuss the epidemiology, clinical presentation, histology, and pathogenesis with review of literatures.

Case Report

A 57-year-old male patient visited our tertiary referral hospital with a chief complaint of a firm mass over the tongue for the proceding 6 months. He had no pain, swallowing difficulty, and neck lymph node enlargement. Physical examination by bimanual palpation revealed a well-defined,

small, spherical, erythematous, firm mass on the dorsal surface of the tongue, with a 1 cm diameter, and the overlying mucosa was slightly raised but intact(Fig. 1). The physical examinations of the other sites were unremarkable. There was no specific past medical history and family history. And there were no previous trauma and inflammations in this area.

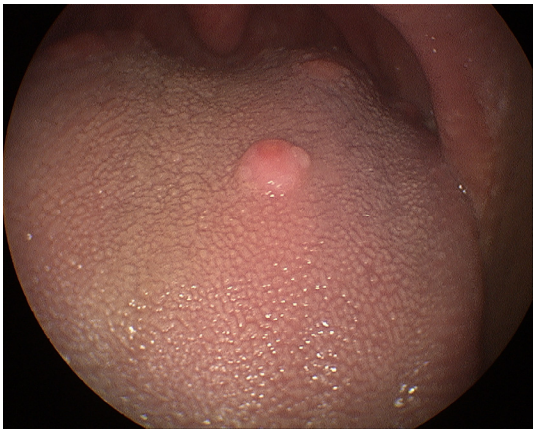

Fig. 1. Preoperative endoscopic finding shows a round, sessile mass in dorsal surface of the tongue

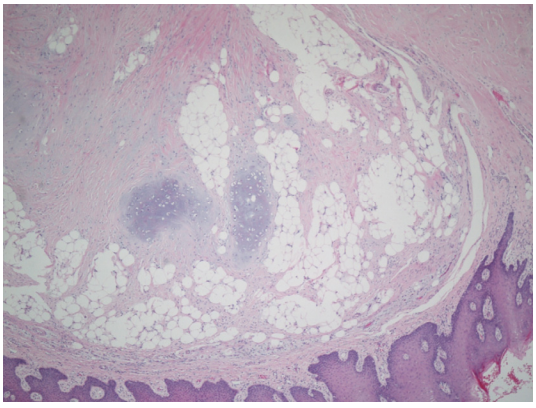

Fig. 2. The photomicrograph shows a well-circumscribed nodule composed of mature hyaline cartilage with typical chondrocyte covered with non-keratinizing stratified squamous cell epithelium (H&E, X40)

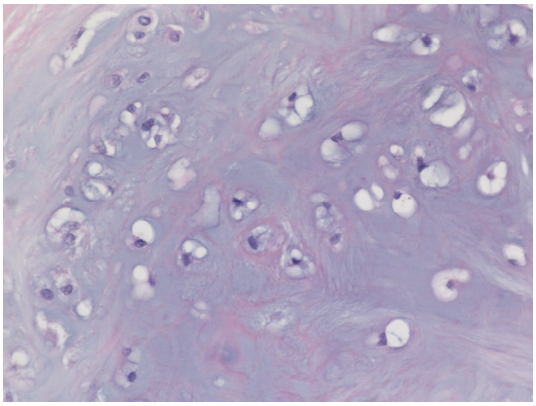

Fig. 3. The photomicrograph shows small chondrocytes with a clear cytoplasm and round nuclei (H&E, X400)

The initial clinical diagnosis was pleomorphic adenoma, granuloma, and choristoma. Surgical excision was performed under local anaesthesia with proper margin. Surgical specimens were sent to pathologists and stained with H&E. This is composed of a piece of nodular mass, measuring 1.1 X 0.6 X 0.5 cm in size. The microscopic study of the tumor showed lobules of well-formed hyaline cartilage and surrounding fibrous tissue in the submucosa. There is no cellular atypia or abnormal mitosis(Fig. 2 and 3). Immunohistochemical stains revealed that chondrocytes and mature fat tissue show strong positive immunoreaction for S-100 protein. Whereas, there is no immunoreaction for cytokeratin and desmin, very low proliferating activity in Ki-67 stain(Fig. 4).

Wound healing was satisfactory, and vicryl wound sutures were removed a week later. The postoperative course showed no recurrence and complication for 6 months.

Discussion

Oral cavity choristomas are extremely rare disease and mainly develop at foramen cecum.[6] In the previous study, 39 cases of choristomas of the oral cavity were reported. Foramen cecum and circumvallate papillae were the most common occurred site (36 cases) followed by the alveolar process and buccal vestibule (3 cases).[1] In the past, choristoma was known as osteoma or chondroma. However, since 1913, 59 cases of choristomas were stated.[7]

Although the cause of disease occurrence is uncertain and controversial, some hypotheses try to describe its origin. First, embryologically tongue anterior two thirds is originated from first branchial arch and posterior one third is originated from third branchial arch. Subsequently, tongue is combined around foramen cecum and probably branchial apparatus multipotency stem cell is regarded as origin of choristoma.[8] Second, thyroid descends from foramen cecum during developmental process and undescended thyroid tissue could be origin of choristoma.[8] Third, posterior one third of tongue is constantly stimulated by tongue movement caused by swallowing, pronunciation and such stimulations lead to calcium deposit and tissue hypertrophy.[9]

Females occurred three to four times more than males and more than half of patients were twenties and thirties.[8] Clinical symptoms of choristomas are globus sensation, dys-

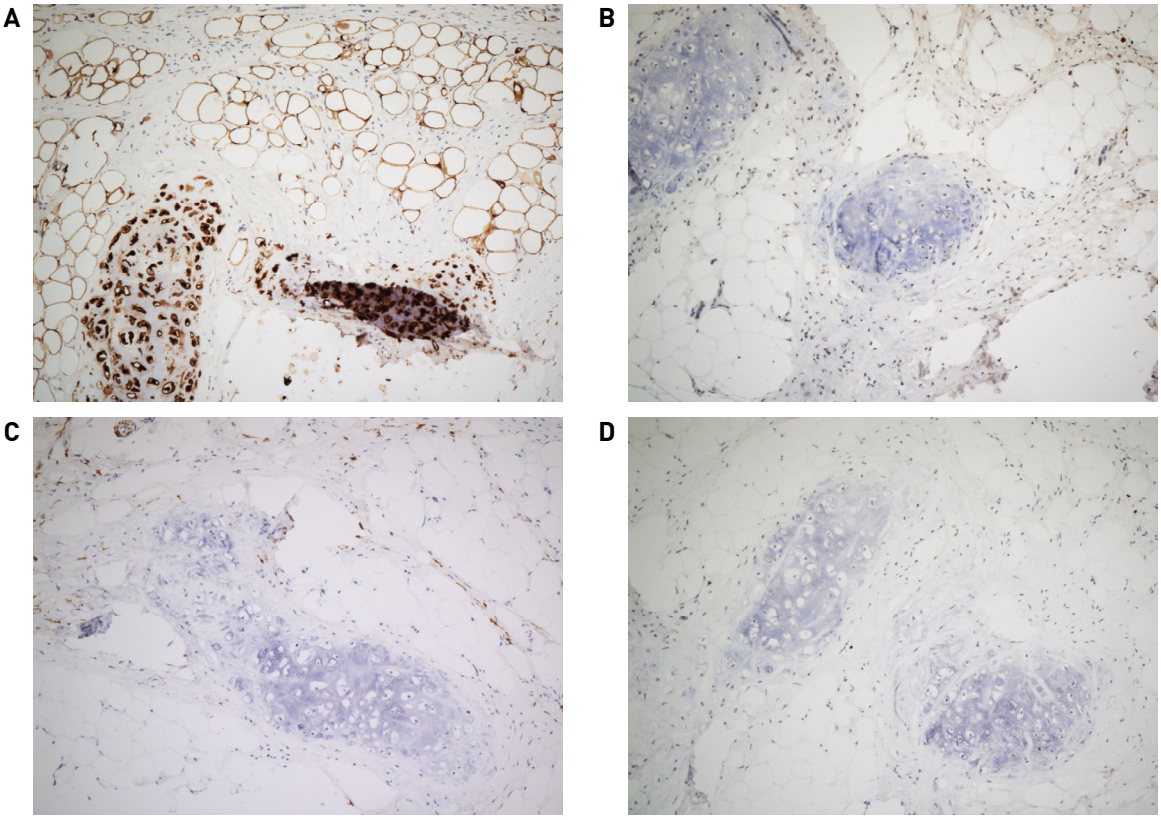

Fig. 4. The immunohistochemical stains of chondroid choristoma. The cartilage lobules are strong immunoreactive to S-100 protein (A, X100). Cytokeratin (epithelial marker, B, X100) and Desmin (C, X100) are not expressed in cartilage lobules. There is very low proliferating activity in Ki-67 stain (D, X100).

phagia, nausea, throat irritability, and snoring. However, symptom-free is most common.[10] The diameter of tumor had a wide range from 4 to 25mm. But, patient's symptoms were not correlation with its diameter. Previous study suggested in patient with largest diameter 25 mm tumor had no symptom, whereas in patient with 7 mm tumor had globus sensation in throat.[11]

Computed tomography scan analysis may be used to check tumor site and its relation to the anatomic structures to exactly plan surgery extent and well defined, round, radio-opaque, and calcification lesion is detected.

Histological examination confirms definite diagnosis of choristomas. Histologically, choristoma is composed of cartilaginous tissue surrounded by fibrous connective tissue with normal chondroblasts without atypia, mitosis, and necrosis.[12] In previous study conducted by Park et al, the microscopic findings of choristoma located at lingual tonsil were consisted of mature bone tissue, covered with non-keratinizing stratifies squamous cell epithelium.[15] By comparison, this case showed lobules of well-formed hyaline cartilage and surrounding fibrous tissue in the submucosa. In this regard, it can be found that chondroid choristoma has various histopathological appearance such as mature hyaline cartilage, bone and fat tissue. Chondroid choristoma resembles benign chondroma histologically, however, choristoma developed in locations that generally do not contain chondrocytes.

In addition, immunohistochemical study is crucial to rule out other disease. Choristoma has high positive S-100 protein (marker of myoepithelial cell) immunoreactivity owing to cartilaginous tissue. There is a negative immunoreaction for cytokeratin (marker of epithelial cell), desmin (marker of muscle cell) and Ki-67 (marker of cellular proliferation).[13] This examination can differentiate choristoma from thyroglossal duct cyst, salivary gland neoplasm, fibroma, lipoma, giant cell tumor, and calcified lymph node.[11] Especially ectopic thyroid gland must be excluded if lesion is near the foramen cecum. Therefore, thyroid function test and thyroid scan must be conducted before surgery when lesion is located at posterior tongue.[14] Chondrosarcoma could be excluded according to cellular and nuclear morphology of the chondrocyte.[11] The present case was simply distinguished

as an encapsulated tumor of mature chondrocytes surrounding by hyaline materials.

The treatment of chondroid choristomas of the tongue is surgical excision. Local recurrence is very rare but incomplete resection of the tumor or surrounding tissue has the possibility to develop new cartilaginous tissue.[1] However, prognosis of choristoma is extremely favorable and long term follow up is not recommended.[1]

In conclusion, oral cavity chondroid choristoma is very rare, but has a good clinical behavior. Surgical excision is a treatment of choice, and post-operative prognosis is extremely good according to the previous study.

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