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성인에서 원주상피를 보이는 설낭 : 증례 보고

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〈Abstract〉

Lingual Cyst with Columnar Epithelium in an Adult : A Case Report

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Lingual cyst is a clinical term indicating a cyst occurring within the tongue. Various disease may clinically present as a lingual cyst, such as foregut cyst, dermoid cyst, thyroglossal duct cyst, salivary duct cyst, hemangioma and ranula. In general, lingual cysts are asymptomatic but may cause airway obstruction, feeding or swallowing problems. Most of these lesions can be simply treated by surgical excision with good postoperative healing and low recurrence rate. Apart from that, histopathological diagnosis of the lesion is important because it means the origin of the lesion. Thus it is important to rule out several origins of lingual cyst histopathologically, because it might be related with adjacent anatomical structure or may indicate cysts on other sites. Herein, we reported a case of lingual cyst with columnar epithelium at the ventral tongue of a 46-year old man and reviewed the clinical and histopathological considerations required for further classification in lingual cysts.

Key words: Lingual cyst, Foregut cyst, Oral foregut cyst, Lingual cyst with columnar epithelium

Ⅰ. INTRODUCTION

The term "Lingual cyst" is a clinical term describing soft tissue cysts arising within the tongue. Lingual cyst is usually asymptomatic but it may cause airway obstruction, feeding or swallowing problems because of its site. When occurred in ventral tongue, elevated tongue might cause severe functional problems and must be excised. Also histopathological examination with clinical consideration are needed to classify the lesion as a specific disease and evaluate its pathologic origin. Various cysts and fluid filled lesions that may be located in the tongue, such as foregut cyst, dermoid cyst, thyroglossal duct cyst, salivary duct cyst, hemangioma and ranula, should be considered for differential diagnosis of lingual cyst. Clinical features, such as lesion location, are helpful in clinically diagnosing lesions like thyroglossal duct cyst which arises in the midline of posterior aspect of the tongue to suprasternal region. Nevertheless, majority of lingual cysts lack specific clinical characteristics and histopathologic evaluation is essential for specific diagnosis. In the tongue, columnar epithelium is present in the interlobular (excretory) ducts of salivary glands, therefore lingual cysts with columnar epithelium is extremely rare. In this case report,

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we will report a case of lingual cyst with columnar epithelium at the ventral tongue and then review the clinical and histopathological considerations required for further classification in this type of lingual cyst..

Ⅱ. CASE REPORT

A 46-year old male patient with ventral tongue swelling and pronunciation difficulty visited our clinic. The patient complained that this discomfort had started a year before the visit. The patient did not have any specific medical or dental history. There was a palpable fluctuating mass located at the midline of the ventral tongue and mid portion of mouth floor. The overlying oral mucosa color and smoothness was within normal range and there were no evidences of surface erosion or ulceration. Magnetic resonance image (MRI) showed an oval shaped benign cystic mass in the deep ventral tongue with a 3.7 x 3.6 x 4.0 cm dimension. (Fig. 1)

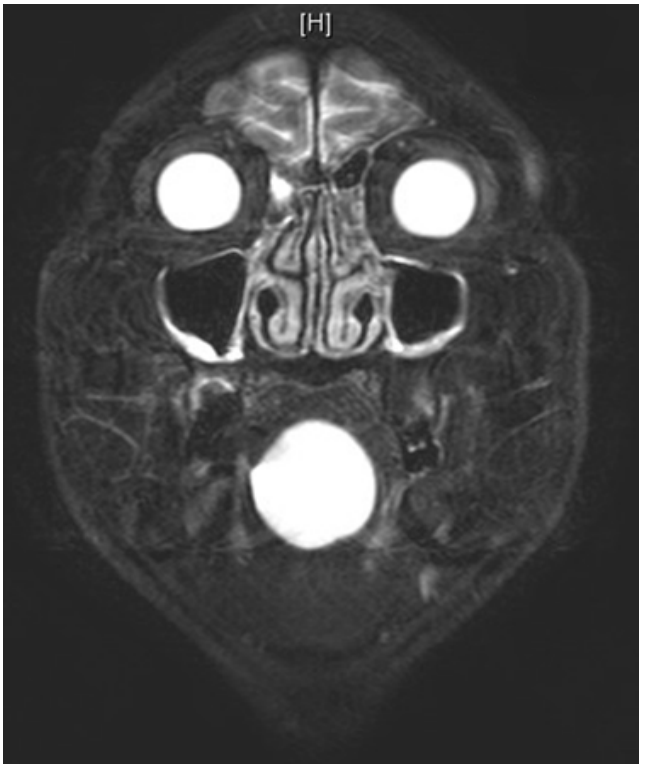

Fig. 1. Magnetic resonance image (MRI) in coronal view (T2-weighted)

Surgical excision was done under general anesthesia with nasoendotracheal intubation. By palpating the cyst, vertical incision was done on the ventral side of tongue. After exposing the cyst, careful dissection was done in order to avoid damage to the cystic wall. The cystic mass was completely excised without tearing. (Fig. 2) The cystic mass was fluctuant and was filled with clear brown fluid. (Fig. 3) The specimen was sent for histopathologic evaluation. 6 months after operation, wound healing was normal and previous discomfort such as poor pronunciation disappeared.

Histopathological examination revealed a mixed cystic epithelium of ciliated columnar epithelium and stratified

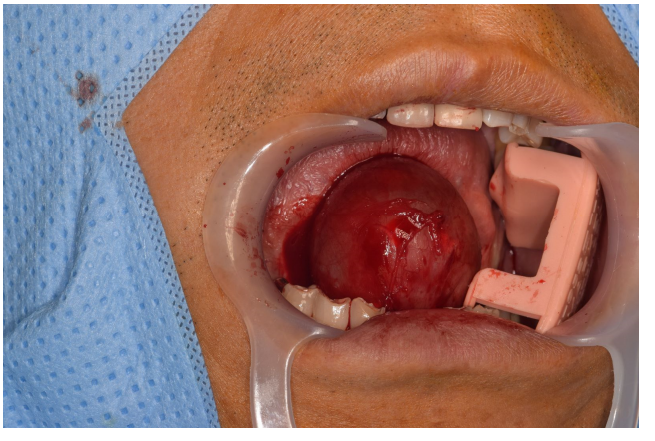

Fig. 2. Clinical image of the cystic lesion after excision

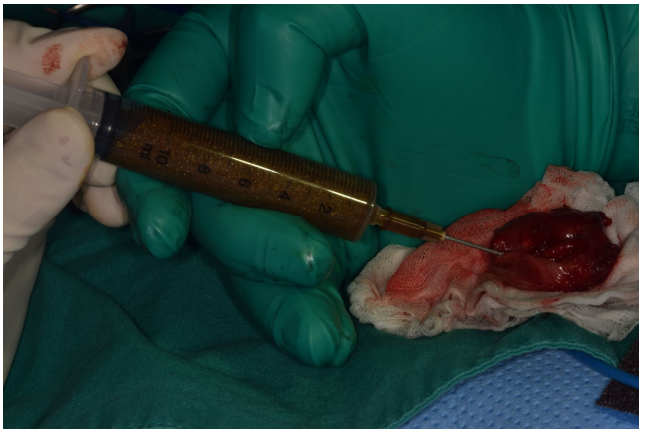

Fig. 3. Cystic lesion filled with clear brown liquid

squamous epithelium. The cystic wall was composed of fibrous connective tissue with chronic inflammation and focal foreign body reaction. (Fig. 4. A, B) Normal adjacent structures such as glossal skeletal muscles were observed at the periphery of the cystic wall. (Fig. 4. C, D) There were no evidence of salivary gland or associated structures. Pathologic differential diagnosis of the specimen was foregut cyst, teratoid cyst or salivary duct cyst due to the cystic epithelium composition. Because of the lack of salivary gland structure and mucus, and the deep intramuscle location of the cyst, salivary duct cyst was ruled out. Foregut cyst of respiratory origin was considered as final diagnosis.

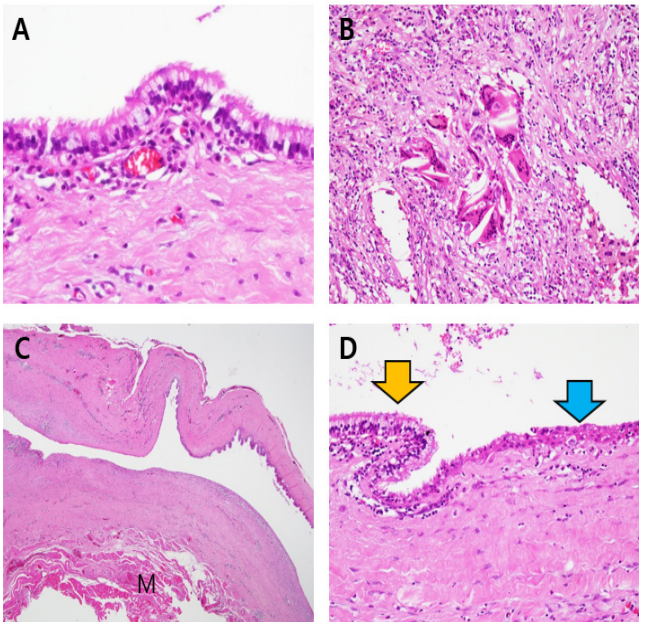

Fig. 4. Histopathologic examination of the surgical specimen. (Hematoxylin and eosin stain)
(A) higher magnification revealed respiratory cystic epithelium and the cyst wall was consisted of fibrovascular tissue (Original magnification x400).
(B) Foreign body giant cells and inflammatory cells were observed in focal areas of the cystic wall, presumably due to rupture of the lesion (Original magnification x200).
(C) The specimen had a cystic structure (lumen and cystic wall) with adjacent glossal skeletal muscle tissue (M) (Original magnification x12.5).
(D) The cyst lining epithelium was consisted mainly of ciliated columnar epithelium (yellow arrow) with focal nonkeratinized stratified squamous epithelium (blue arrow) (Original magnification x200

Ⅲ. DISCUSSION

Both foregut cyst and teratoid cyst may present of columnar and squamous cystic epithelium in lingual cysts. We reviewed and discussed the clinical and histopathological features of the two candidates. The final diagnosis of our case as oral foregut cyst of respiratory origin was made through a comprehensive consideration of clinical and pathologic aspects mentioned henceforth.

1. Foregut cyst

Foregut cyst is a rare choristoma from foregut-derived epithelium that can arise in an ectopic fashion anywhere along the foregut developmental region[1,2]. The lesions has various terms of nomenclature, such as "foregut duplication cyst", "choristoma", "bronchogenic cyst", "enteric cyst", "lingual cyst with respiratory epithelium" or "lingual cyst with gastric epithelium"(in oral lesions), depending on the origin of its epithelium or degree of differentiation[2,3]. Oral foregut cysts are extremely rare, and are most common in the mouth floor and anterior tongue with the majority of the lesions including the midline[2,4,5]. The lesions are most common during infancy and are generally asymptomatic unless they are enlarged or have been infected. There have been several theories for its origin. The most well-known theory is that it has originated and grown from pinched or misplaced embryonic endoderm rests entrapped within the developing tongue in the pharyngeal arch[6,7]. The ventral foregut develops into the respiratory tract, while the dorsal foregut develops into the gastrointestinal tract[7]. Another theory is that it originated from pluripotent embryonic tissue that may differentiate into either respiratory or intestinal epithelium, or both[8]. Foregut cyst has one or more or the following epitheliums; respiratory, intestinal or

squamous epithelium for its cystic lining. An underlying smooth muscle layer is generally required for diagnosis but can be indistinct or absent in lingual foregut cysts with respiratory dominant epithelium[2,4,9]. An attached location to foregut origin has been proposed for foregut cyst diagnosis as well, yet the spatial relation is indistinct in the developed tongue unlike the proximity of the foregut and embryonic developing tongue[4,9] Stromal cartilage and mucous gland formation are possible features[3,4].

2. Dermoid cyst / Epidermoid cyst / Teratoid cyst

Dermoid cyst, epidermoid cyst and teratoid cyst are developmental cysts classified as a benign cystic form of teratoma[10]. In the oral cavity, they are commonly observed at the midline mouth floor and deep tongue and present as a sublingual swelling that may cause dysphagia and dyspnea[11]. Although they are regarded a teratoma, the ectoderm is the dominant germ layer of which the lesion presents its tissue features and tissues derived from other germ layers are extremely rare. Dermoid cysts and epidermoid cysts consist of only 1 germ layer, the epidermis, and are not true teratomas, while teratoid cysts consist of 2 or more germ layers and are more rarer the former two types[12]. Dermoid cyst has an epidermal-like orthokeratinized stratified squamous epithelium as its cystic lining and skin adnexa (sebaceous glands, sweat glands and hair follicles) in the cystic wall, while epidermoid cyst has an identical cystic epithelium without appendages[11]. The presence of rich orthokeratin observed in the cystic lining and cyst cavity is characteristic and gives a thick yellowish appearance at gross examination. Teratoid cysts have been noted to have a distinct keratinized squamous epithelium structure along with a mixture of additional features from the endoderm or mesoderm; bone, muscle, adipose tissue, cartilage, respiratory or gastrointestinal epithelium[11].

There are case reports using the terminology dermoid cyst with respiratory or gastric epithelium[13,14,15], yet the presence of respiratory or gastrointestinal epithelium gives controversies of the epithelium origin as true teratoma or choristoma of foregut origin[16]. Moreover, some of these case reports proposed the cystic origin of their case as the endodermal cell in the foregut which indicated the difficulties in distinct discrimination between teratoid cyst and foregut cyst[15]. Our case had only a focal area of non-keratinized squamous epithelium with dominant ciliated columnar epithelium and did not have adnexal tissues, which seemed insufficient and distinct compared to the previously reported teratoid cyst cases which exhibited definite keratin formation and adnexal structures. The histopathologic characteristics of this case were more consistent with those of a foregut cyst and it was thought to be more reasonable to assume the lesion to be a foregut originated choristoma.

The surgical treatment procedures for foregut cyst and teratoid cyst are both enucleation and they have excellent prognosis. Nevertheless, rare malignant transformation has been reported in oral foregut cysts[17] and demoid/teratoid cysts[18,19]. Malignant transformation in foregut cysts present as adenocarcinoma[20], while dermoid/teratoid cysts result as squamous cell carcinoma and/or sarcoma.

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