

DERMOID CYST ON THE FLOOR OF THE MOUTH : A CASE REPORT

Shan-Shan Liang¹, Yeon-Gyu Chu¹, Chin-Soo Kim¹, Sang-Han Lee¹, Ki-Jeong Byeon²¹*Dept. of Oral & Maxillofacial Surgery, School of Dentistry, Kyungpook National University*²*Dept. of Dentistry, College of Medicine, Ulsan University***Abstract**

Dermoid cysts located in the floor of the mouth are very rare developmental keratinizing squamous epithelium lined cysts. Anatomically, they are classified as sublingual (median genioglossal), submental (median geniohyoid) and lateral dermoid cysts, and they can be further classified as epidermoid, dermoid, and teratoid cysts by histology. We report a case of sublingual dermoid cyst in a 16-year-old boy presenting as a large sublingual swelling causing speech and swallowing difficulties and discuss the surgical treatment techniques and histopathological features of this lesion.

Key words : Dermoid cyst, Floor of mouth, Sublingual dermoid cyst

I . Introduction

Dermoid cysts located in the floor of the mouth are very rare cystic malformations lined with keratinizing squamous epithelium. They occur primarily in the testes and ovaries. Head and neck dermoid cysts take less than 10% of all dermoid cysts^{1,2}. The most common location in the head and neck is the lateral third of the eye brow, and the second most common location in the head and neck region is the floor of the mouth^{3,4}, which take 23%-34% of all head and neck and account for only 1.6%-6.5% of all body dermoid cysts³⁻⁷. Depending on their location, dermoid cysts on the floor of the mouth are divided into medial and lateral cysts. In adults, they are termed as sublingual or genioglossal cysts when located between the geniohyoid and mylohyoid muscles, when located between the mylohyoid and the neck cutaneous muscles, they are termed as submental or geniohyoid cysts. Therefore, they are classified as sublingual (median genioglossal), submental (median geniohyoid), and lateral dermoid cysts according to the anatomic relationship between their location and the muscles of the floor of the mouth. Histologically, they can be further classified as epidermoid, dermoid or teratoid according to their overlying epithelium

and the contents inside the cavity⁵. Though dermoid cysts represent a separate entity, the term "dermoid" is generally used to indicate all three categories.

These cysts generally present as slowly and progressively growing lesions, and even if they are congenital, a diagnosis is normally made in the second or third decade of life⁸. Only few cases have been described in childhood. Recommended treatment is surgical excision via intraoral or extraoral access, depending on the lesion's size and location^{9,10}.

In this report, we presented a case of 16-year-old boy with a large sublingual dermoid cyst occupying the entire floor of the mouth, and we successfully excised it by employing intraoral access with a transverse mucosal incision and achieved good cosmetic and functional results with no significant complications.

II . Case Report

A 16-year-old boy sought treatment in the dept. of OMFS, Kyungpook National University Dental Hospital for mouth floor swelling that had existed for about 7 years, although he had not noted any significant increase in size over these last 7 years. He is allergic to flower powder, though he doesn't contact

flower powder recently, the size of the lesion wasn't noted to be decreased significantly. He described mild discomfort in swallowing and speaking, but well tolerated.

Examination revealed a rubbery, painless swelling underneath the tongue, adhered to the deep plane, displacing his tongue upwards, and unfluctuant on palpation. However, nontender, and no lymphadenopathy was observed. The appearance of the mucosa of the mouth floor is normal (Fig. 1). The size and appearance of the orifices of Wharton's ducts are normal. The larynx was normal. The tumouration can't be palpated at the submentonian level and there are no laterocervical adenopathies.

Panoramic radiograph did not reveal any pathology. CT scan revealed a midline, hypodense, unilocular, well defined capsule cystic mass without calcification, and peripheral enhancement was revealed by contrast-enhanced CT, but central enhancement within the cyst is unusual, and the contents may have slightly negative attenuation values(Fig. 2). The cyst was located between the geniohyoid and the mylohy-

oid muscles and corresponds to a sublingual (median genioglossal) cyst. The preliminary diagnosis was a sublingual dermoid cyst.

Surgical excision was performed with an intraoral approach by a transverse mucosal incision (Fig. 3) over the sublingual mass under general anesthesia delivered by nasotracheal intubation. A mouth retractor was positioned to keep the mouth open, and a retraction suture was placed on the tip of the tongue. A transverse mucosal incision was made over the sublingual mass. Submucosal dissection was made delivering a well encapsulated mass from the sublingual area. No dense attachment to neighboring tissues was detected. The patient remained intubated for 12 hours postoperatively because of concern for tongue swelling. His postoperative course was uneventful.

The specimen was egg-like shape, measuring 6×5×4 cm, the extent of the lesion as demonstrated by CT scans was confirmed (Fig. 4). The outer surface of the cyst was smooth and regular and the cyst lumen contained a yellow sebaceous material. Tissue



Fig. 1. An unfluctuating tumouration was observed, painless and displacing his tongue upwards. The appearance of the mucosa on the floor of the mouth is normal. The size and appearance of the orifices of Wharton's ducts are normal.

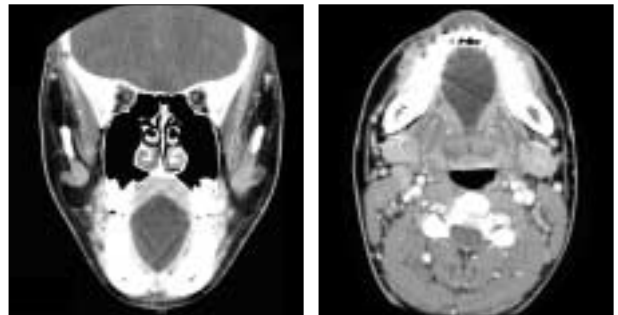


Fig. 2. CT(coronal and axial views), cystic tumouration of the floor of the mouth, well delimited.



Fig. 3. Exeresis of the cyst by intraoral transverse mucosal approach.



Fig. 4. Image of the cyst egg-like shape, measuring 6×5×4cm, the outer surface was smooth, regular.

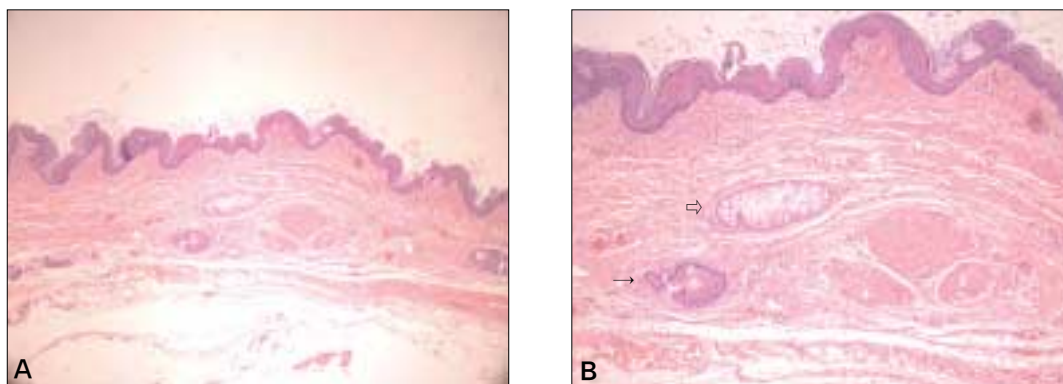


Fig. 5. A : The section shows dermoid appendages are present in the cyst wall, which lined with stratified squamous epithelium. (HE stain, $\times 40$). B : Note the cyst lined by keratinized stratified squamous epithelium with dermoid appendages containing sebaceous glands (\rightarrow) and hair follicle (\Rightarrow) (HE stain, $\times 100$).

samples were stained with HE. Final histopathologic result showed the cyst lined by stratified squamous epithelium with sebaceous glands and hair follicle, which confirmed the diagnosis of a dermoid cyst (Fig. 5).

III. Discussion

Dermoid cysts derived from pluripotential cells of ectodermal origin are very rare developmental keratinizing squamous epithelium lined cysts. The pathogenesis of the cysts of the floor of the mouth is not well established, but theories of dysontogenetic, traumatic, and thyroglossal duct anomaly have been suggested⁸.

Histologically, all true dermoid cysts are lined by epidermis with the presence of adnexae such as sweat glands, sebaceous glands, hair and hair follicles. If no adnexa presents, the entity is termed as epidermoid cyst. If there are structures in their cyst walls derived from all three germinal layers (such as nervous, gastrointestinal, and respiratory tissues) with the lining cells varying from stratified squamous to a ciliated respiratory epithelium, then the entity is called teratoma or teratoid cyst.

In the present case, CT typically demonstrated a midline hypodense unilocular mass with a well defined capsule locating between the geniohyoid and the mylohyoid muscles, which was benefit for the surgeon to choose the most appropriate surgical approach, especially for very large lesions.

Complete surgical excision was the treatment of choice. Several techniques were reported in the literature, which may be divided into intraoral and extraoral access depending on the lesion's size and location. Regarding intraoral approach, a midline vertical mucosal incision along the ventral surface of the tongue¹¹ was used for small sublingual cysts ; a bilateral incision along the mandibular ridge crest was described by Lowry et al¹² ; a median glossotomy technique was used for median genioglossal (sublingual) cysts ; an extended median glossotomy technique was used for very large median genioglossal (sublingual) cysts. The latter two techniques allowed to obtain a very good approach to the cyst and to obtain adequate surgical control of the lesion in the event of median cysts located above the geniohyoid muscles⁸. In addition, a symphyseal mandibular osteotomy was introduced by McGregor¹³ to enucleate a very large sublingual dermoid cyst. The extraoral approach was generally preferred in the case of submental cysts or very large sublingual cysts, whereas the intraoral approach was typically used for smaller sublingual cysts. However, Akao et al¹⁴ reported a case of large dermoid cyst of the floor of the mouth, and they stated that intraoral access must be attempted first, even if dealing with a large cyst. In our case, the cyst was so large and we wanted to attempt the intraoral approach, so we chose a transverse mucosal incision, by which we successfully excised the large cyst and achieved good cosmetic and functional results.

The prognosis was very good without major complications, but it should be kept in mind the operation by intraoral access may damage structures in the sublingual space and may ensure hemorrhage and hematoma formation, leading to potentially life threatening complications. Modest edema was the most common postoperative complication encountered, with little alteration in tongue movement, swallowing, and breathing. Pryor et al¹⁵⁾ described that only 1 patient had recurrence following up 49 pediatric dermoid cysts patients of the head and neck, but not in the floor of the mouth. To the best of our knowledge, no recurrence case at the floor of the mouth following a complete surgical excision has been reported until now. Malignant transformation to squamous cell carcinoma has rarely been reported, but not in the floor of the mouth.

Conclusively, we successfully excised a large sublingual dermoid cyst by employing intraoral access with a transverse mucosal incision and achieved good cosmetic and functional results. In our opinion, the intraoral approach is effective in the treatment of large sublingual dermoid cyst at the floor of the mouth, for that it leads to very good cosmetic and functional results. The extraoral incision is mandatory only when the cyst lies under the geniohyoid muscle.

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저자 연락처

우편번호 700-412
대구광역시 중구 삼덕동2가 188-1
경북대학교 치의학전문대학원 구강악안면외과학교실
김진수

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Chin-Soo Kim
Dept. of OMFs, School of Dentistry, Kyungpook National University
188-1, Samduck-dong 2-ga, Jung-gu, Daegu, Korea
Tel: 82-53-600-7551
E-mail: kimcs@knu.ac.kr

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