

Sublingual Dermoid Cyst in a Female Infant

— With Special Reference to Reported Cases in Japan —

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INTRODUCTION

It is well known that dermoid and epidermoid cyst originate either from a malformed germinal epithelium inclusion occurring during the prenatal period or epithelium implantation caused by trauma or infection¹⁻⁵. And otherwise there is teratoid cyst^{3,4,6-9}. Such cysts may occur in every part of the human body, and approximately 1.6% of all dermoid cysts only arise in the mouth floor, corresponding to the anatomical site of the III group by the classification of Mayo Clinic⁴. Even though asymptomatic in infancy, most such lesions prefer to become symptomatic in young adults aged 15 to 35^{1,2,5}. Sublingual dermoid cyst in an infant, which is regarded as a teratoid type, is rare. This paper aims to report such a case and to consider clinicopathologically with special reference to reported cases in Japan.

CASE REPORT

Patient: 11-months-old female baby.

Date of first examination: September 30th, 1986.

Chief complaint: Lump formation of the mouth floor.

Previous history: The patient's mother first noticed the tumor when the baby was 9-months of age. At 11 months, the baby was taken with to our hospital. There was no prominent change in size during that period. Physical condition was good.

Present status: The tumor, 3.5×2 cm in size, was

located at the midline region of the mouth floor and its margin was obvious. The surface was smooth and round. It was movable and elastic soft. No fluctuation was palpated, but sticky-milky content was aspirated. The tumor caused abnormal lifting-up of the tongue (Fig. 1-A), but motor disturbance (sucking disorder) was likely absent, according to the mother's statement.

Clinical diagnosis: Probable dermoid or epidermoid cyst

Laboratory examination: non-pathologic

Surgery and postoperative course: By the intraoral approach under GOF general anesthesia, the cyst was easily enucleated en bloc without adhesion to Wharton's ducts (Fig. 1-B). The postoperative course was uneventful.

Gross specimen: The outer and inner surfaces were smooth and uniform (Fig. 2-A). The cavity, filled with a milky porridge-like substance, contained no hairs (Fig. 2-B).

Histopathology: The cyst wall covered by epithelial lining of squamous cells with various degrees of keratinization (Fig. 3-A, B). Desquamated keratin was seen in the cyst cavity (Fig. 3-B). Ciliated and goblet cells were also observed (Fig. 3-A, C). The cyst wall had skin appendages such as hair root and sebaceous glands in addition to mesodermal components (Fig. 3-B).

Histologic diagnosis: Dermoid cyst

DISCUSSION

Only 6.9% of dermoid and epidermoid cysts occur in the oral region⁴. The oral region is not a prominent predilection site for occurrence of these cysts, with the mouth floor even less prominent. Mouth floor cysts are classified into two types by location as follows; Sublingual type is located between the oral mucosa and geniohyoid mus-

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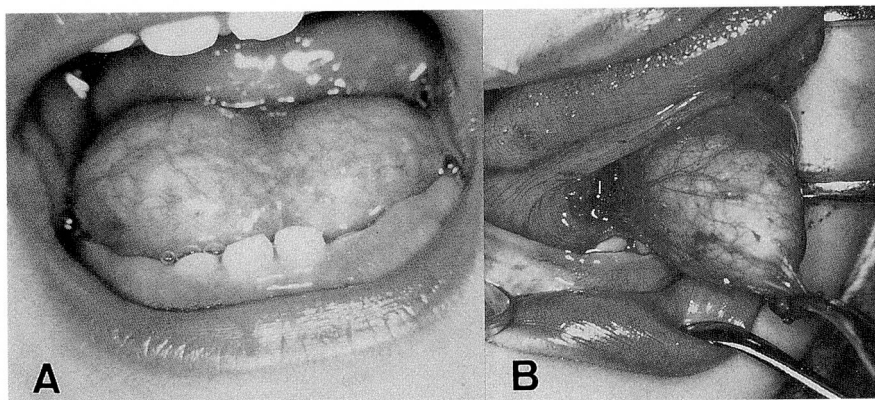


Fig. 1-A Preoperative oral appearance showing lifting-up of the tongue
1-B Gross appearance of the cyst during enucleation



Fig. 2-A The specimen
2-B The enucleated cyst with porridge-like content

cle, and submental type arises between geniohyoid muscle and the skin³⁾. Our case was one of the sublingual type. The cysts of the mouth floor are distinguished into two groups by occurrence site as follows; one is the midline type, the other is the lateral type. Most mouth floor cysts are of the midline type³⁾. Seward's detailed description¹⁰⁾ of occurrence site subclassification is very interesting, but the debate is better abbreviated here.

In the literature published in Japan from 1936 to 1988^{9,11)}, 19 cases of the oral region in infants were able to be collected including our case. Of these, 17 cases were occurred in the mouth floor (Table 1). Cyst size was usually several centimeters across on average (variable from soybean to hen's egg in size). Our case belonged to a larger group.

In most of these lesions, symptoms occur after becoming relatively young adults aged 15 to 30. Cases showing

symptoms are relatively rare in infants^{9,12)}.

The most prominent symptom of the sublingual type is sucking disturbance in case of large cysts. Though the cyst of our case was large, fortunately there was no such a disturbance. Based on the present pertinent literature, sex difference, age, histologic type and location type were summed up in Table 1.

Judging from histopathologic classification, epidermoid cyst is listed prior to dermoid cyst in incidence. As patient's age increased, epidermoid cyst occurred more often than dermoid cyst did. The interrelated results between of histologic findings and age suggest that infant case might be different from adult case in etiology and/or in the time course of cyst development. Of these 10 cases, many cyst walls histologically had skin appendages such as sebaceous glands and sweat glands. In a few cysts, hair and hair follicles were also seen. The cyst

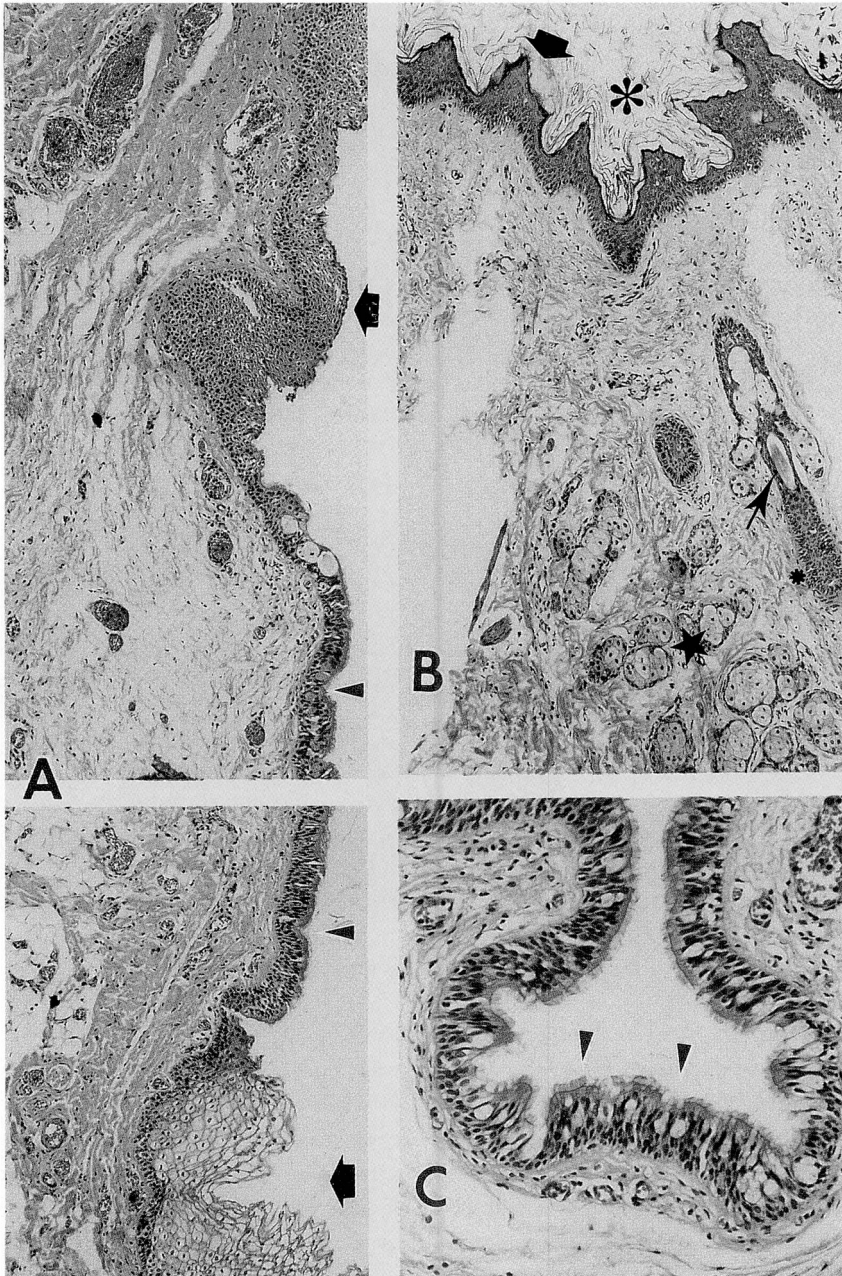


Fig. 3-A The cyst wall covered by various epithelial linings with squamous cells (◄) and ciliated columnar cells (◄) (H & E, $\times 50$)
3-B Keratinizing squamous cell (◄), keratin squames (*), skin appendages like hair (●), hair follicle (*) and sebaceous gland (★) (H & E, $\times 50$)
3-C Magnification of ciliated columnar cell and goblet cell (H & E, $\times 200$)

wall of our case was lined by stratified squamous epithelium with skin appendages, although complete hairs were not contained in the cyst cavity. Kunimi¹²⁾ reports that a

dermoid cyst may histologically change into an epidermoid cyst. This report suggest that these skin appendages in the cyst wall might have gradually become atrophied and

Table 1 Analyses of mouth floor cysts in infancy from the Japanese literature

sex	male	7	
	female	8	<2>
age	at birth	10	
	within one week	1	<1>
	after one week	5	
histology	dermoid	10 (M:F=6:3)	<1>
	epidermoid	6 (M:F=1:4)	<1>
	teratoid	1	
location	sublingual type	11	
	submental type	2	<4>

< >: undescribed

retrograded. Therefore, it may phenomenally agree that dermoid cysts occur frequently in childhood and epidermoid cysts increase in young adulthood. By the way, the cyst wall of the present case possessed had ciliated columnar cell, goblet cell and distinct mesodermal components in addition to dermal appendages, suggesting a teratoid cyst^{6,8)}. There are still many unsolved clinicopathologic problems concerning these cysts.

Complete surgical removal should be recommended for treatment. The prognosis of our case was excellent.

SUMMARY

A case is presented of congenital dermoid cyst occurring at the mouth floor of a 11-months-old female baby. The cyst cavity was mainly lined by squamous epithelia with skin appendages such as hair follicles and sebaceous glands. Also seen were ciliated columnar cells and goblet cells in some parts of the cyst wall. The coexistence of respiratory-typed epithelium might suggest that this case belonged to a teratoid type. Such cases of the mouth floor were clinicopathologically considered with special

reference to the pertinent literature in Japan.

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