A Benign Cystic Teratoma in the Floor of the Mouth of an Infant

—A Case Report—

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Summary: One case of a benign cystic teratoma on the bottom of the oral cavity of an infant was observed and reported. The occurrence of this tumor in the oral cavity is quite unusual.

Key words: benign cystic teratoma—oral cavity—infant—the floor of the mouth—dermoid cyst

Introduction

A benign cystic teratoma is a cystoma with tridermic components frequently occurring in the ovary, testicle, mediastinum and orbit, but rarely observed in the oral cavity (Anderson and Kissan, 1977). Recently the authors observed such a tumor in an infant, which is described below.

Case Report

A 25-day old male infant was referred to the oral surgery department on February 7, 1977, for a swelling in the floor of his mouth.

History

The patient was born on January 12, 1977 at a public hospital by normal delivery. His weight was 4,140 g, height 52 cm, measurements around the head and the chest 36.2 cm and 35.5 cm, respectively, at birth. Three days after birth a tumor was noted at a median location on the bottom of the oral cavity. It was suspected to be a thyroglossal cyst, however, there was no difficulty in breathing and suckling. Upon discharge he was referred to this department. There was no abnormality observed during the pregnancy.

Clinical examination

Except for colored skin from jaundice of the newborn, nothing was abnormal. There was no cyanosis nor dyspnea.

A hemispherical tumor, 2.5 cm in diameter, was in the floor of the mouth and it elevated the tongue (Fig. 1). It was soft on palpation, and no fluid wave was palpable. With exploratory puncture, about 1 ml of a viscous fluid with a white turbidity was suctioned.

Laboratory studies

The laboratory studies were performed at the time of admission. The results were as follows: leukocyte count, 9600/mm³; erythrocyte count 4,140,000/mm³; platelet, 188,000/mm³; hemoglobin concentration, 11.2 g per 100 ml.; liver function, normal; Wasserman serum test, negative.

Treatment and Prognosis: there was no special dysfunction, but at the patient's convenience a follow-up observation was
Fig. 1. Photograph showing a mass in the floor of the mouth at first examination.

Fig. 2. Photograph taken at admission.
BENIGN CYSTIC TERATOMA OF THE MOUTH

Fig. 3. Surgical specimen.

Fig. 4. Cut surface of surgical specimen.
Fig. 5. Photomicrograph showing a cyst wall of the tumor composed of keratinizing, stratified squamous epithelium and pseudostratified ciliated epithelium with mucous glands and striated muscle in the connective tissue.

Fig. 6. Photomicrograph showing a cyst wall of the tumor containing hair follicles, sebaceous glands and sweat glands.
performed. At the 9th month after birth the tumor began growing (Fig. 2), thus on October 31 of the same year the patient admitted, and on November 4th the tumor was extirpated under general anesthesia.

An incision was made in the mucosa at the midline of the floor of the mouth, above the tumor, extending from the ventral aspect of the tongue to the linguogingiva. After blunt dissection of the surrounding tissue, the tumor was found to extend across both mandibles up to the mylohyoid muscle where it formed a cord, and passed through the inside of the mylohyoid muscle to the hyoid bone. The tumor was completely enucleated as a unit. The postoperative course was without complication. On the 7th postoperative day, the patient was discharged. Three years later, there was no sign of recurrence and he remained asymptomatic. Histopathology-Grossly, the specimen had a fig-shape with a smooth surface, that was pink and somewhat yellowish. It weighed 9g and measured 3.5×2.5×2.5 cm (Fig. 3). The cut surface revealed a thin-walled, cystic structure, containing a white, foamy, turbid, viscous and creamy substance. The wall was 2 to 3 mm thick, the inside of which was smooth at the globe and coarse at the cord (Fig. 4). Microscopically, the globular part consisted of stratified squamous epithelium and the cord consisted of pseudostratified ciliated epithelium. In the lower layer of squamous epithelium, dermatophyte tissue such as hair follicles, sebaceous glands and sweat glands were observed, and in the lower layer of ciliated epithelium, mucous glands and striated muscles were seen (Fig. 5 and 6). From these clinical and pathological findings it was diagnosed as a benign cystic teratoma.

Discussion

A cystic teratoma is considered to be a cystic tumor having tridermic components originating from totipotential cells scattered promiscuously in a tumor. Since the present tumor had tridermatic components, namely, ectodermal tissue with dermatophytes such as sweat glands, sebaceous glands and hair follicles, mesodermal tissue with striated muscle, and endodermal tissue with mucous glands and respiratory epithelium, it can be classified as a benign cystic teratoma. This type of tumor generally occurs in the ovaries and testicles, occasionally in the mediastinum and orbit, but no report is available describing this tumor in the oral cavity.

It can be assumed that this tumor rarely occurs in the oral cavity, and that it is reported as a type of dermoid cyst. As pointed out by Thoma (Gorlin and Goldman, 1970), the term dermoid cyst has been erroneously used to describe a benign cystic teratoma of the ovary for a long time.

Meyer (1955) described the dysontogenetic cysts occurring on the bottom of the oral cavity, which can be classified histopathologically into three types: (1) epidermoid type, (2) dermoid type and (3) teratoid type. Generally this classification is used and the tumor in the present case was a teratoid type. When statistics relating to dermoid or epidermoid cysts in the oral cavity area were studied, Takahashi and Onishi (1952) found no teratoid types in 73 cases of stomatic dermoids which were reported in the domestic literature published for 30 years prior to 1952. Tamari and Mawatari (1968) collected 62 reports of dermoid-epidermoid cysts around the oral cavity from domestic and foreign cases published from 1957 to 1967. Only one stomatic case of the teratoid type was noted in Japan in a 26-year-old male by Takakita and Takahashi (1963) and an-
other stomatic case was reported overseas by Meyer (1955). Furthermore, Enrin et al. (1976) reported on a collection of 135 cases of dermoid cysts around the oral cavity from domestic reports published from 1968 to 1975, including one case of the teratoid type, although the details are not known.

On the other hand, Colp (1925) reported on stomatic dermoid cysts in 3 of his own patients in addition to 32 cases collected from other reports. No teratoid type cysts were observed.

New and Erich (1937) reported in detail on 1,495 cases of dermoid cysts treated in the Mayo Clinic from 1910 to 1935. According to the report 103 cases involved the head and neck, of which 24 involved the stomatic area (on the bottom of oral cavity, inframentum, infragnathia). None were the teratoid type.

Yoshimura et al. (1970) described 8 cases of dermoid cysts in the area of the oral cavity including some of their own patients and some cases found in the domestic and foreign literature from 1949 to 1967. Four cases were reported to be of the teratoid type. One case by Meyer (1955), one case each by Miller and Owens (1966) on the tongue, and one case by Quinn and Robinson (1965) on the bottom of oral cavity were noted.

However, the case of Quinn and Robinson (1965), as well as Bury’s (1962), can hardly be the teratoid type with no tridermic components, and they should be included with the 15 cases of cysts with gastric and intestinal mucosa reported by Gorlin and Tirasek (1970) as heterotic cysts.

Thus, the occurrence of dermoid cysts of the teratoid type in the area of oral cavity is very rare. Only the four cases by Mayer (1955), Orsos et al. (1956), Takakita and Takahashi (1963) on the bottom of oral cavity and Miller and Owens (1966) on the tongue have been described.

Many studies on the clinical pathology of this tumor in the ovary are available. According to Marcial-Rojas and Medina (1958) the percentages of ectodermal-, mesodermal- and endodermal tissues in 268 cases with this tumor are 100%, 92% and 72%, respectively. Ectodermal tissue including dermatophyes such as subaceous glands and hair follicles, and stratified squamous epithelium were observed in nearly all the cases. Cerebral tissue and nerve fibers were also seen in 41% and 38% of the cases, respectively. Mesodermal tissue including muscle is seen in as many as 92% of the cases followed by a 35% occurrence of bone and a 22%

### TABLE 1

Reported cases of benign cystic teratoma in the oral cavity

<table>
<thead>
<tr>
<th>author</th>
<th>sex</th>
<th>age</th>
<th>location</th>
<th>squamous epithelium</th>
<th>sebaceous glands</th>
<th>hair follicles</th>
<th>sweat glands</th>
<th>striated muscle</th>
<th>bone</th>
<th>fat</th>
<th>mucous glands</th>
<th>gastrointestinal epithelium</th>
<th>respiratory epithelium</th>
</tr>
</thead>
<tbody>
<tr>
<td>Meyer (1955)</td>
<td>M</td>
<td>newborn</td>
<td>floor of the mouth</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Orsos (1956)</td>
<td>F</td>
<td>10 years</td>
<td>floor of the mouth</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
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<td></td>
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</tr>
<tr>
<td>Kokita (1963)</td>
<td>M</td>
<td>26 years</td>
<td>floor of the mouth</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
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</tr>
<tr>
<td>Miller (1966)</td>
<td>F</td>
<td>newborn</td>
<td>tongue</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td></td>
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</tr>
<tr>
<td>Kameyama (1960)</td>
<td>M</td>
<td>newborn</td>
<td>median floor of the mouth</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
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occurrence of cartilage. Furthermore, endodermal tissue had a low incidence of 72%, which includes mucous glands, gastrointestinal mucosa and respiratory epithelium, among which respiratory epithelium was the highest at an incidence of 48%. Rarely thyroid tissue is seen (3%).

Table 1 summarizes the reports on the tumors in the area of the oral cavity. Four cases were found on the bottom of the oral cavity, one case on the tongue and 3 cases in infants. In the histological studies cerebral tissue, tooth and thyroid tissue were not observed unlike the ovary.

In the ovary this tumor may become malignant; but in the stomatic area few examples are available, thus no malignant cases have been observed. The present case has shown no sign of recurrence, years after surgery and the course is favorable; however the patient must be followed carefully in the future.

References


