Title: Congenital Epidermoid Cyst Arising in Soft Palate Near Uvula: A Case Report

Author(s): Suga, K; Muramatsu, K; Uchiyama, T; Takano, N; Shibahara, T

Journal: Bulletin of Tokyo Dental College, 51(4): 207-211

URL: http://hdl.handle.net/10130/2132
Case Report

Congenital Epidermoid Cyst Arising in Soft Palate Near Uvula: A Case Report

Kenichiro Suga, Kyotaro Muramatsu, Takeshi Uchiyama, Nobuo Takano and Takahiko Shibahara

Department of Oral and Maxillofacial Surgery, Tokyo Dental College, 1-2-2 Masago, Mihama-ku, Chiba 261-8502, Japan

Received 12 May, 2010/Accepted for publication 21 June, 2010

Abstract

Aberrant ectodermal tissues during the fetal period or acquired aberrant epithelial tissue due to trauma or surgery are thought to cause dermoid and epidermoid cysts. Their incidence is 7.0% in the head and neck region and only 1.6% in the oral cavity, where they mostly present in the floor of the mouth. On the other hand, they are extremely rare in the soft palate and uvula, and only six cases have been reported. Epidermoid cysts grow slowly and asymptptomatically, and thus rarely cause oral dysfunction. However, cysts arising in the floor of the mouth can lead to developmental disorders due to impaired sucking and swallowing. This report describes a 4-week-old boy in whom an epidermoid cyst developed in the midline region of the soft palate close to the uvula. Decreased sucking ability led to a poor in body weight, so resection was performed at an early age. The histopathological diagnosis was epidermoid cyst.

Key words: Epidermoid cyst—Congenital cyst—Soft palate—Uvula—Neonate

Introduction

Dermoid and epidermoid cysts are thought to be caused by aberrant epithelial components of ectodermal tissues during the fetal period or by aberrant epithelial tissue acquired due to trauma and surgery. Such cysts can arise anywhere in the body, but the incidence in the head and neck region accounts for only 7.0% of the total. Among cysts of the head and neck, incidence in the stomatognathic region is highest in the floor of the mouth and lowest in the soft palate. To the best of our knowledge, only six reports have described cysts involving the uvula or soft palate. Cysts often develop slowly and asymptptomatically, but when they arise in the floor of the mouth of infants, respiration and feeding disorders can result.

We describe herein a neonate in whom suckling was decreased due to a congenital epidermoid cyst in the soft palate.

Case

A 4-week-old boy who was delivered normally in September 2004 was brought to our
Fig. 1 Intraoral condition at first examination
Revealed small, whitish, domed mass under mucous membrane.

Fig. 2 Intraoral condition during surgery
a: Mass was located in medial soft palate, behind uvula (arrows).
b: Border between mass and surrounding tissue was clear (arrows).

Fig. 3 Excised lesion
Spherical cystic lesion measured $7 \times 8 \times 8$ mm.

Fig. 4 Histopathological findings
Cystic wall was lined with flat, stratified squamous epithelium with slight keratinization; keratinous materials were evident inside cyst. Epidermoid cyst was diagnosed.
hospital for an intraoral medical check-up at the age of 4 weeks. He appeared to have developed normally, and his nutritional status was favorable. A soft elastic, soybean-shaped mass was observed in the soft palate at the base of the left side of the uvula. The extremely thin oral mucosa covering the mass allowed visual confirmation of a white hemispherical mass beneath (Fig. 1). The lesion was asymptomatic, and because suckling volume and behavior appeared normal and no clinical malignancies were evident, the patient was only monitored. However, at the age of 8 weeks, the lesion had slightly increased and the patient had gained insufficient weight due to poor suckling ability. A further examination led to a diagnosis of a benign soft palate tumor, and resection was scheduled.

Surgery proceeded under general anesthesia with oral intubation in accordance with benign tumor resection procedures. The surgical field was brightened using a Dingman mouth gag, and the lesion could be visualized from the posterior base of the uvula to the posterior edge of the soft palate. The uvula was pulled using a nylon suture, and mucosal resection included the mucosa covering the mass along the base of the mucosa (Fig. 2-a). The border between the mass and surrounding tissue was clear, which facilitated its separation and resection from the surrounding tissue (Fig. 2-b). Because a deep part of the mass reached the muscle coat of the tensor veli palati and levator veli palatini muscles, each muscle bundle was sutured after resecting the mass and the surgical wound was closed by suturing. Since his systemic condition stabilized and suckling improved postoperatively, the patient was discharged in good health seven days later. The patient has remained free of recurrence for five years.

The excised spherical mass measured about $7 \times 8 \times 8$ mm, and the surface was flat and smooth (Fig. 3). A histopathological examination showed that connective tissue had formed a cystic cavity covered by keratinized stratified squamous epithelium. The cyst wall was slightly keratinized and was lined with keratinized stratified squamous epithelium, and the cyst itself contained keratinous materials (Fig. 4). No evidence of dermal appendages presented in the cyst wall. Based on the above findings, the lesion was diagnosed as an epidermoid cyst.

Table 1: Published cases of dermoid/epidermoid cyst of palate or uvula

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year</th>
<th>Age</th>
<th>Gender</th>
<th>Site</th>
<th>Size (cm)</th>
<th>Symptoms</th>
<th>Treatment</th>
<th>Histopathological diagnosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>Green and Neal</td>
<td>1982</td>
<td>18yr</td>
<td>F</td>
<td>midline of soft palate</td>
<td>unknown</td>
<td>unknown</td>
<td>surgery</td>
<td>dermoid cyst</td>
</tr>
<tr>
<td>Pruszewicz et al.</td>
<td>1984</td>
<td>neonate</td>
<td>unknown</td>
<td>uvula</td>
<td>unknown</td>
<td>decrease of suckling</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td>Yoshinari and Nagayama</td>
<td>1986</td>
<td>12 mts</td>
<td>F</td>
<td>uvula</td>
<td>0.6×0.4</td>
<td>asymptom</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td>Zappia et al.</td>
<td>1991</td>
<td>62yr</td>
<td>M</td>
<td>soft palate</td>
<td>3.5</td>
<td>hypopomolence, snoring</td>
<td>aspiration, surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td>Seshul et al.</td>
<td>1998</td>
<td>1. 12 mts</td>
<td>M</td>
<td>uvula</td>
<td>1.0×1.0</td>
<td>snoring</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. 12 mts</td>
<td>M</td>
<td>uvula</td>
<td>0.5</td>
<td>dysphagia</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td></td>
<td></td>
<td>3. 3 mts</td>
<td>M</td>
<td>uvula</td>
<td>0.5</td>
<td>asymptom</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td>Mu et al.</td>
<td>2002</td>
<td>1. neonate</td>
<td>M</td>
<td>uvula</td>
<td>0.4×0.4</td>
<td>asymptom</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
<tr>
<td></td>
<td></td>
<td>2. 1 mth</td>
<td>F</td>
<td>uvula</td>
<td>0.5×0.5</td>
<td>asymptom</td>
<td>spontaneous rupture</td>
<td>epidermoid</td>
</tr>
<tr>
<td>present case</td>
<td></td>
<td>1 mth</td>
<td>M</td>
<td>midline of soft palate</td>
<td>0.7×0.8×0.8</td>
<td>decrease of suckling</td>
<td>surgery</td>
<td>epidermoid</td>
</tr>
</tbody>
</table>


Discussion

Dermoid and epidermoid cysts can arise anywhere in the body, and according to the study of New and Erich\(^3\), the most common location was the anus (44.5%) followed by the ovary (42.1%) in 1,495 patients. The incidence in the head and neck region was 6.9%, which accounted for only a small fraction of the total. Furthermore, of 103 patients with head and neck dermoid and epidermoid cysts, the most common locations were the orbit (46.6%), followed by the floor of the mouth and submental region (23.3%), nose (12.6%), neck (10.7%) and lip (2.9%). Only six dermoid and epidermoid cysts in the soft palate or uvula have been reported (Table 1).

Acquired factors include aberrant epithelial components in deep tissue due to trauma, and inflammation and surgical complications may be mechanisms of onset, but dermoid and epidermoid cysts are usually congenital. The most frequent congenital cause is aberrant ectoderm in the fusion area of the branchial arch and groove at the fetal age of 3–4 weeks, but cleaved cells can be ectopically reactivated during the fetal period\(^3\).

In stomatognathic areas other than the palate, most cysts arise in the midline region where the left and right branchial arches fuse. Several investigations have localized such cysts arising in the soft palate or uvula in the midline region. The secondary palate that comprises the palatine and soft palate is formed when the left and right palatine shelves of the maxillary process fuse at the fetal age of around 7 to 9 weeks, supporting the notion that epidermoid cysts arising in the soft palate or uvula are due to congenital causes involving aberrant ectoderm in the fused area. The mass in the present patient extended slightly laterally from the midline region, and it is impossible to ascertain which congenital factor(s) was involved in the cyst. However, the mass was confirmed soon after birth, and the cyst was already present at the age of one month. Therefore, congenital factor(s) were indicated by the absence of trauma in this case. The basic treatment for epidermoid cysts is total excision. Ronald\(^6\) treats extremely large masses via fine-needle aspiration to reduce the content and then performs the excision. However when the objective of such surgery is unclear, palliative treatments such as fine-needle aspiration and fenestration can lead to infection, which potentially causes pain and exacerbation\(^4\). Therefore fine-needle aspiration should be performed as a diagnostic tool, and total excision should be promptly performed if epidermoid cysts are diagnosed clinically. While various theories have been proposed for the timing of surgery for these cysts, surgery for neonates should be performed when safe to do so under general anesthesia. The findings in the present patient did not indicate malignancy the mass was quite small at first examination and dysfunction was absent. Hence we originally planned to wait for the patient to mature before proceeding with more thorough testing. However, because suckling had become mildly impaired by the age of 8 weeks, we decided to resect the mass sooner than planned under strict perioperative management. The results in this case indicate that surgery performed at an early age without fine-needle aspiration helped facilitate a marked improvement in suckling and increase in body weight.

References


Reprint requests to:
Dr. Kenichiro Suga
Department of Oral and Maxillofacial Surgery,
Tokyo Dental College,
1-2-2 Masago, Mihama-ku,
Chiba 261-8502, Japan