Pediatric Intraoral Ranulas: An Analysis of Nine Cases

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YUCA, K., BAYRAM, İ., ÇANKAYA, H., ÇAKSEN, H., KIROĞLU, A.F. and KİRİŞ, M. Pediatric Intraoral Ranulas: An Analysis of Nine Cases. Tohoku J. Exp. Med., 2005, 205 (2), 151-155 — An intraoral ranula is a retention cyst arises from the sublingual gland on the floor of the mouth as a result of ductal obstruction and fluid retention. Many techniques for management of ranulas have been described in the literature. The purpose of this study was to analyze our surgically treated pediatric patients with intraoral ranulas and to discuss the results in the light of the literature. Nine pediatric patients (six females and three males) with intraoral ranulas surgically treated were analyzed retrospectively regarding their treatment methods and results. The surgical specimens were also re-examined histologically. Seven cases of superficial, protruded and smaller than 2 cm ranulas were treated with marsupialization (unroofing). Two cases who were previously operated and then recurred had bigger than 2 cm ranulas. In these two cases, marsupialization of the ranula plus removal of the sublingual gland was performed. The most common complication was intraoperative cyst rupture of the ranula, which was noted in four cases. A recurrence was observed in only one case in the 16th months of follow up period. Our findings show that marsupialization is a suitable and effective method for pediatric intraoral ranulas, whereas in recurrent cases marsupialization of the ranula combined with total excision of sublingual gland may be preferred ——— ranula; intraoral cyst; sublingual mass; marsupialization

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The term “ranula” is derived from the Latin word “rana” (meaning frog) and is descriptive of the bluish, slow enlargement of the floor of the mouth to form a painless, fluctuant, translucent, dome-shaped swelling, which is said to resemble the underbelly of a frog (Urso-Baiarda et al. 2003; Haberal et al. 2004). An intraoral ranula is a retention cyst arises from the sublingual gland on the floor of the mouth as a result of ductal obstruction and fluid retention (Urso-Baiarda et al. 2003). The etiology is unknown but it has been described in association with congenital anomalies, trauma, and disease of the sublingual gland (Davison et al. 1998). An important feature in the histologic diagnosis is the absence of epithelial tissue in the pseudocyst wall (Davison et al. 1998).

In this study nine children with intraoral ranulas surgically treated were analyzed retrospectively. Our purpose was to analyze our surgically treated pediatric patients with intraoral ranulas.

Received March 16, 2004; revision accepted for publication December 10, 2004.
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Unfortunately, we could not get any information about their previous surgical intervention. In all cases, ranulas were sublingual type. Seven cases of superficial, protruded and smaller than 2 cm ranulas were treated with marsupialization (unroofing). Two cases who were previously operated and then recurred had bigger than 2 cm ranulas. In these two cases, marsupialization of the

MATERIALS AND METHODS
The study included nine pediatric patients (six females, and three males) with intraoral ranulas of which seven were superficial and smaller than 2 cm and two were bigger than 2 cm. All cases were surgically treated in Yüzüncü Yıl University Faculty of Medicine, Department of Otorhinolaryngology between 1996 and 2003. The patients’ data were retrospectively investigated on the basis of their medical records. In our study, superficial ranulas smaller than 2 cm were treated with only marsupialization (unroofing), but ranulas recurred and bigger than 2 cm were treated with marsupialization plus removing of the sublingual gland. The surgical specimens were re-examined histologically by a pathologist. The study was approved by the ethics committee of our university hospital.

RESULTS
Our study consisted of six females and three males with intraoral ranulas treated surgically. The patients’ age ranged from 7 to 15 years (mean 10.8 ± 2.66 years). The mean time between the awareness of the lesion to admittance was 2.88 ± 2.89 months (1-9 months).

In four cases, ranulas were on the right side, in the others on the left (Fig. 1). Seven cases were surgically treated in our clinic, whereas two cases were previously operated in another medical center and presented to our clinic for recurrence.

Fig. 1. Gross appearance of intraoral ranula: Intraoral ranula on the floor of the mouth.

Fig. 2. Ranula: Predominance of histiocytes in the cystic space and on the pseudocystic fibrous connective wall is shown (Hematoxylin and Eosin stain, original magnification × 200, scale bar: 200 μm).

Fig. 3. Ranula: Central cystic space and a wall composed of loose, vascularized connective tissue and sublingual gland are demonstrated (Hematoxylin and Eosin stain, original magnification × 50, scale bar: 1 mm).
ranula plus removing of the sublingual gland was performed. Postoperative packing of the entire ranula with gauze for two days was performed in all cases.

Histopathological findings showed predominance of histiocytes in the cystic space and on the pseudocystic fibrous connective wall (Fig. 2). Central cystic space and a wall composed of loose, vascularized connective tissue and sublingual gland are shown in Fig. 3.

As a complication intraperative cyst rupture of the intraoral ranula was noted in four cases. A recurrence was noted in the 16th month of follow-up in case 2. This patient was treated with marsupialization of ranula and removal of the sublingual gland. The clinical findings of the patients are summarized in Table 1.

### DISCUSSION

Ranula is a clinical term generally used for cystic lesions in the floor of the mouth (Quick and Lowell 1977; Morita et al. 2003). There are two different concepts in the pathogenesis of ranula. One is a true cyst formation due to ductal obstruction with an epithelial lining, and the other is a pseudocyst formation due to ductal injury and extravasation of mucus without an epithelial lining (Quick and Lowell 1977; Urso-Baiarda et al. 2003). Ranula may also uncommonly present as a rapidly enlargement and swelling following infection (Urso-Baiarda et al. 2003). In our series only one case (case 4) had a history of rapidly enlargement of the lesion following viral upper respiratory infection.

Aside from ranula, a number of other lesions may be encountered in the floor of the mouth or submandibular space region. These include congenital abnormalities (cystic hygromas, branchial cleft cysts, and thyroglossal duct cysts), benign lesions (epidermoid cysts, dermoid tumors, and lipomas), malignant neoplasia, and other lesions (abscess, mucocele, and sarcoidosis) (Urso-Baiarda et al. 2003). While the most of the mentioned lesions required detailed investigations the diagnosis of ranula is largely clinical (Morita et al. 2003; Urso-Baiarda et al. 2003). In our study, the diagnosis of ranula was confirmed by histopathologically in all of our cases.

Treatment of an intraoral ranula consists of surgical excision. Excision of the sublingual gland is considered to be a reasonable and suitable option (or intervention) for radical treatment (Haberal et al. 2004). Marsupialization (unroofing) is also reported to be effective surgical procedure. With the simple addition of the packing of the entire ranula cavity with gauze after marsupialization, the rate of recurrence is minimized (Baumash 1992; Haberal et al. 2004).

### Table 1. Characteristics of our pediatric with ranula

<table>
<thead>
<tr>
<th>Case number</th>
<th>Sex</th>
<th>Age (years)</th>
<th>Side of ranula</th>
<th>Time between awareness of the lesion and admittance (months)</th>
<th>Operation type</th>
<th>Complications</th>
<th>Treatment of recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Female</td>
<td>12</td>
<td>Left</td>
<td>1</td>
<td>MP</td>
<td>Cyst rupture</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>Female</td>
<td>10</td>
<td>Right</td>
<td>2</td>
<td>MP</td>
<td>-</td>
<td>MP + SE</td>
</tr>
<tr>
<td>3</td>
<td>Male</td>
<td>14</td>
<td>Right</td>
<td>4</td>
<td>MP</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>4</td>
<td>Male</td>
<td>11</td>
<td>Right</td>
<td>1</td>
<td>MP</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>Female</td>
<td>9</td>
<td>Left</td>
<td>1</td>
<td>MP + SE</td>
<td>Cyst rupture</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>Female</td>
<td>15</td>
<td>Right</td>
<td>9</td>
<td>MP</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Female</td>
<td>7</td>
<td>Left</td>
<td>6</td>
<td>MP</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>Male</td>
<td>12</td>
<td>Left</td>
<td>1</td>
<td>MP</td>
<td>Cyst rupture</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>Female</td>
<td>8</td>
<td>Left</td>
<td>1</td>
<td>MP + SE</td>
<td>Cyst rupture</td>
<td></td>
</tr>
</tbody>
</table>

MP, Marsupialization; SE, Sublingual gland excision.
et al. (1988) reported that the incidence of recurrence after conventional marsupialization of ranulas or pseudocysts of the oral floor was in the range of 61-89%. Therefore, they recommended that lesions larger than 1 cm should be treated by excision of the sublingual gland. Bridger et al. (1989) suggested that this treatment method might be applied to all ranulas regardless of their size. In our study, seven cases of superficial, protruded and smaller than 2 cm ranulas were treated with marsupialization. The cases previously operated and then recurred were treated with removal of the sublingual gland combined with marsupialization of the ranula. Postoperative packing of the entire ranula with gauze for two days was performed in all cases.

It has been reported that structures at risk during ranula excision include submandibular duct and lingual nerve (Urso-Baiarda et al. 2003; Haberal et al. 2004). Among complications, we observed only intraoperative cyst rupture of the intraoral ranula.

Choi and Oh (2003) used hydrodissection technique in the treatment of ranulas and they noted less bleeding, fewer incidents of neural and soft-tissue damage, and a lower recurrence rate. They also reported that their hydrodissection technique is relatively simple, effective, and convenient for otolaryngologists. This procedure involves the injection under pressure of saline and lidocaine with 1:100,000 of epinephrine into the dissection plane. Removing of a ranula without causing rupture can be a tedious and time-consuming process (Choi and Oh 2003).

Watanabe et al. (2002) and Fukase et al. (2003) reported that intracystic injection therapy with OK-432 is relatively safe and can be used as a substitute for surgery in the treatment of ranulas. Alternatively, the ranula can be treated with the placement of a silk suture or seton into the dome of the cyst (Morton and Bartley 1995; Haberal et al. 2004). Pandit and Park (2002) recommended that optimal management of pediatric oral cavity ranulas may include observation for 5 months for spontaneous resolution. If the lesion does not resolve or recurs repeatedly, surgical treatment is recommended (Pandit and Park 2002).

Many patients do not come to control examination after discharging from the hospital because of low socioeconomic levels of the population in our region. Therefore, we could not permit to spontaneous resolution in our cases. However, we also agree with Pandit and Park (2002) about waiting for spontaneous resolution of intraoral ranulas. If the lesion does not resolve or recur, surgical treatment can be recommended.

Haberal et al. (2004) suggested that marsupialization should firstly be tried in pediatric population. Takagi et al. (2003) described a new method of fenestration and continuous pressure as a simple, effective and non-invasive procedure for the treatment of plunging ranulas. We also agree with Haberal et al. (2004) because our seven cases of superficial ranulas were treated with marsupialization. We suggests that superficial and smaller than 2 cm ranulas may be treated with marsupialization and packing.

A controversy is whether the rupture of the cyst increases the risk of recurrence (Haberal et al. 2004). In our study rupture of cysts was noted in four cases, and only one case recurred.

In conclusion, our findings have shown that marsupialization is a suitable and effective method for pediatric intraoral ranulas, whereas in recurrent cases marsupialization of the ranula combined with total excision of sublingual gland may be preferred.

References


