



## Are the patients with anatomic variation of the sublingual/Wharton's duct system predisposed to ranula formation?



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### ABSTRACT

**Objective:** To evaluate ranula development according to anatomic variation of the ductal system of sublingual gland (SLG), especially the presence of Bartholin's duct.

**Methods:** The anatomic variation of SLG duct was prospectively investigated and compared between 55 consecutive patients with ranulas treated by SLG excision (group 1) and another 15 consecutive patients undergoing similar surgeries for other conditions (group 2). The ductal structures of SLGs and submandibular glands (SMG) were also compared between the pediatric patients and adult patients with ranulas.

**Results:** In 32 of 55 patients with ranulas (58.2%) and 1 of 15 patients without ranulas (6.7%), the SLG showed an anatomic variation of the main duct called Bartholin's duct structure ( $P < 0.01$ ). Seventeen of 22 (77.3%) pediatric patients with ranulas had Bartholin's ducts and 15 of 33 (45.5%) adult patients with ranulas had Bartholin's ducts ( $0.01 < P < 0.05$ ), but Bartholin's duct which opens near to the orifice of Wharton's duct was not found in this study population. Plunging ranulas that extravasate deep to the mylohyoid were observed in the 6 patients (4 children and 2 adults). There was no recurrence in all cases with ranulas.

**Conclusions:** Congenital anatomic variation of the ductal system of the SLG might be a possible cause of ranulas in the patients with simple ranulas, especially in pediatric patients. Surgical resection of the SLG is a better treatment choice for ranulas than other conservative treatments.

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### 1. Introduction

Ranula is a mucus pseudocyst (not lined by the epithelium) resulting from the retention or extravasation of saliva from the sublingual gland (SLG) in the floor of the mouth or in the neck region [1], common in the second decade of life [2]. It is described as a blue, translucent swelling in the floor of the mouth, so is said to resemble the throat of a croaking frog [3].

The two types of ranulas are the simple (intraoral) ranula and the plunging (cervical) ranula according to their extent [1].

The reliability of a diagnosis of intraoral ranula is principally based on clinical signs and imaging [1], but sometimes, ultrasonography [4], CT, MRI [4], and fine needle aspiration (FNA) [5] have been used for the diagnosis of a plunging ranula particularly in pediatric patients. Many methods of treatment for ranulas have been reported in previous studies and with different results [1,4,6–12]. The usual treatment of a ranula is marsupialization [1] and the preferred treatment for recurrent or persistent ranulas is excision of the ranula as well as the SLG via an intraoral approach to the anterior of the floor of the mouth. However, the total SLG excision is associated with a certain risk of complications. These include injury to the lingual nerve or Wharton's duct, with possible stenosis, leading to obstructive sialadenitis and ductal laceration, causing salivary leakage [12,13].

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The precise etiology of ranulas remains unproven, but may associate with local trauma or previous surgery [1], disease of the SLG [1], and congenital anomalies [14–16]. The oral and plunging ranulas are cystic extravasation mucocoeles that arise from the SLG and usually from a torn duct of Rivinus [4]. A congenital predisposition has been suggested, given the predominance of plunging ranulas in people of Asian origin [16]. There is an unusually high incidence of pediatric plunging ranula in some ethnic groups, such as Maoris and Pacific Islanders [17,18]. Infant ranula is rare, but several cases have been reported in the literature [6,19,20]. The local trauma or previous surgery and disease of the SLG are also rare in pediatric patients, especially in infant patients. During meticulous dissection and resection of the SLG for the treatment of ranulas, a considerable number of SLGs have showed the anatomic variation of Bartholin's duct emptying into Wharton's duct.

Based on the above situation, we speculated that congenital anatomic variation of the SLG ductal system, especially the presence of Bartholin's duct, may be a possible cause of ranulas, results in ranula development. In this study, we observed the ductal structures of SLG and submandibular gland (SMG) in 70 surgical procedures and analyzed the relationship between ranulas and anatomic variation of the SLG ductal system for supporting our hypothesis.

## 2. Patients and methods

### 2.1. Patients

This prospective study was conducted on 55 consecutive cases of ranulas in 55 patients treated by SLG excision (Group 1; age from 2 to 57 years old, mean 17.5 years old) in the period from January 2004 to August 2014. The main criterion was that the patients with a history of ranular surgery were excluded from this study.

We also observed the ductal structure of the SLG and the SMG in 15 patients without ranulas (age from 1 to 70 years old, mean 45.1 years old) from January 2010 to December 2014, necessitating dissection of the FOM, including cases of mouth floor cancer (10 cases), cylindroma (2 cases), myoepithelioma (1 case), epidermoid cyst (1 case), and chronic sialinguitis (1 case). These 15 cases without ranulas formed a control group (Group 2).

The patients with ranulas in this study were subdivided into pediatric group (younger than 14 years old) and adult group (older than 14 years old) and the relationship between ranulas and anatomic variation of the SLG ductal system was analyzed in these two groups.

All cases were collected from the department of oral and maxillofacial surgery of the Hunan Provincial People's Hospital. The diagnosis was based on clinical features, and ultrasonography, CT, MRI, and FNA (based on aspiration of mucous, presence of amylase in the fluid) which were used for differential diagnosis, particularly in the patients with plunging ranulas and the final diagnosis was confirmed after histologic evaluation of the surgical specimens.

This study was approved by the Institutional Review Board of Hunan Provincial People's Hospital, and informed consents were obtained from the parents of the minor patients and agreed to the adult patients.

### 2.2. Methods

In our study population, all patients with simple ranulas underwent excision of the involved SLG with intraoral removal of the ranula, and other patients with plunging ranulas underwent intraoral excision of the involved SLG with or without removal of plunging ranula, but the pseudocyst was drained and its cervical

contents were evacuated through the intraoral incision. In Group 1, all pediatric patients were subjected to surgical procedures under general anesthesia with oral endotracheal intubation or nasotracheal, and most of adult patients were under general anesthesia with nasotracheal intubation, only 5 adult cases under local anesthesia. In Group 2, all cases were under general anesthesia with nasotracheal intubation. All of the operations were performed by 1 group comprising a senior surgeon (M.B.Z.) and 2 residents.

In the process of operation, a Denhardt mouth gag was introduced for opening the patient's mouth at the contra-lateral side to the ranula. A linear incision was made in the floor of the mouth for all patients, parallel to and just lateral to the submandibular duct. An electrocautery was used for incision of the floor of the mouth distal to Wharton's duct and this duct was identified with meticulous dissection by wearing a loupe. The lingual nerve was also identified to avoid injury. The SLG was freed from Wharton's duct and mucosa using a cold instrument. The contents of the pseudocyst were drained during the dissection of ranula or after removal of the SLG. In the cases with Bartholin's duct variation, Bartholin's duct emptying into Wharton's duct was ligated with 3–0 silk sutures and safely divided from Wharton's duct. Otherwise, the saliva of the SMG might accumulate on the floor of the mouth through the remnant branch connected to Wharton's duct after surgery. Following removal of the SLG, hemostasis was obtained and the mucosa of the floor of the mouth was loosely closed with two or three resorbable sutures to protect the lingual nerve and Wharton's duct. In some cases with plunging ranulas, the drain with rubber strip was placed to prevent a collection of blood or saliva and removed after 48 h. All patients with ranulas were contacted by telephone and followed-up for at least 1 year.

In the control cases (Group 2), for fear of an oncological risk, the examination of the ductal structures of SLG and SMG was carefully performed in the surgical specimens after operation.

### 2.3. Statistical analysis

The relationship between ranulas and anatomic variation of the SLG ductal system was evaluated by Fisher's exact test. The data analysis and statistical test were performed using SPSS version 19.0 (SPSS, Chicago, IL). The *P* values less than 0.05 were considered statistically significant.

## 3. Results

The demographic data and clinical characteristics of ranula are summarized in Table 1. The mean age of the patients with ranulas

**Table 1**  
Demographic, clinical characteristics of 55 patients with ranulas.

	No. of patients (n=55)
Age (years)	17.5 (range 2–57)
Sex	
Male	18 (32.7%)
Female	37 (67.3%)
Location	
Right	22 (40%)
Left	32 (58.2%)
Bilateral	1 (1.8%)
Type of ranula	
Oral	49 (89.1%)
Plunging	6 (10.9%)
Size (mm)	27.3 (range 20–60)

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