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Case report

A rare case of plunging ranula with local recurrence and wide spread to the infratemporal fossa treated successfully by an intraoral surgical treatment

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ABSTRACT

A ranula is a salivary gland cyst that typically presents as a localized superficial swelling on the floor of the mouth. A plunging ranula may develop when extravasation of mucus extends through or around the mylohyoid muscle and deeper into the neck, and can present as a neck lump with or without swelling on the oral floor. Here, we report a case of repeated local recurrence of a plunging ranula that expanded into the infratemporal fossa and was successfully treated by an intraoral surgical treatment. An 11-year-old otherwise healthy Japanese girl presented with extravasation and swelling in the left submandibular region, decreased flow of saliva from the sublingual and submandibular glands, and a 3 × 2 cm fluctuant swelling on the oral floor. Magnetic resonance imaging showed a large plunging ranula expanding to the infratemporal fossa. Surgical fenestration of the cyst wall and extirpation of the sublingual gland was performed using an intraoral approach. A Penrose drain was placed in the oral floor after fenestration of the cyst wall. The lesion was found to have expanded from the submandibular space to the infratemporal fossa through the parapharyngeal space. Drainage was effective, and the postoperative course was uneventful and without recurrence after 1.5 years of follow-up.

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1. Introduction

A ranula is a salivary gland cyst that typically presents as a localized superficial swelling on the floor of the mouth. A plunging ranula may develop when extravasation of mucus extends through or around the mylohyoid muscle and deeper into the neck, and can present as a neck lump with or without swelling on the oral floor [1]. Here we report on a patient with persistent local recurrence of a plunging ranula that had expanded to the infratemporal fossa and was treated successfully by definite surgical treatment using an intraoral approach. Expansion of a ranula from the pterygomandibular space into the parapharyngeal space is unusual, and expansion further to the infratemporal fossa, as in our case, is extremely rare [2]. A plunging ranula is likely to recur, so it is nec-

essary to have a treatment plan that takes into consideration the patient's circumstances, in particular age and the extent of expansion of the lesion.

2. Case report

An 11-year-old Japanese girl was referred to our department in April 2015 with a chief complaint of swelling on the left side of the oral floor and submandibular region. Two years earlier, she had twice undergone fenestration of a cyst on her oral floor at other local clinics of Otorhinolaryngology. She was otherwise in good general health. Extravasation and swelling in the left submandibular region were noted on extraoral examination (Fig. 1). Intraoral examination revealed decreased flow of saliva from the sublingual and submandibular glands and a 3 × 2 cm fluctuant swelling on the floor of the mouth.

Magnetic resonance imaging (MRI) showed a cystic lesion of low and high density cystic nature of spreading mass seen on both T1 and T2 weighted images respectively, located in the left sublingual and submandibular and parapharyngeal space and further expanding to the infratemporal fossa from the oral floor (Fig. 2). Clinical laboratory data were within normal limits. The differential diagnosis

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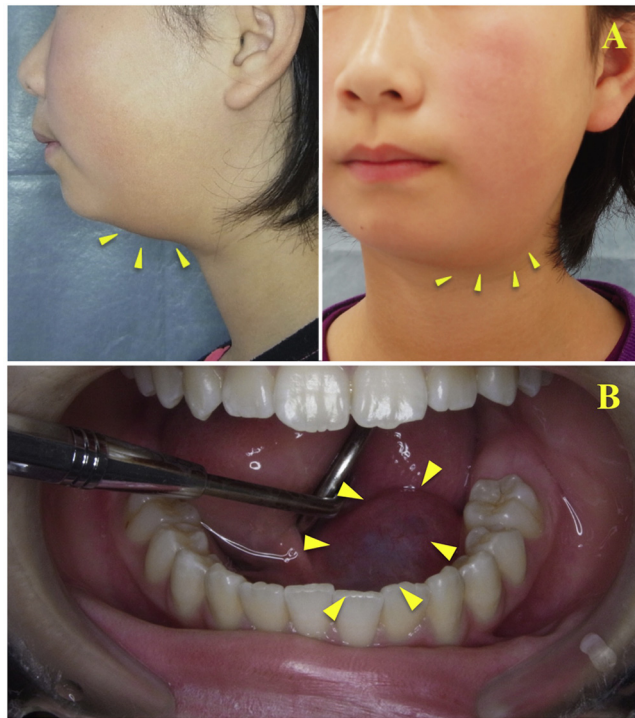


Fig. 1. Intraoral and extraoral photographs. (A) Swelling of the left submandibular region (arrow heads). (B) A painless 3 × 2 cm lesion covered with a normal mucosal surface of colored translucent appearance on the oral floor (arrow heads).

should be considered includes cystic lymphangioma, epithelioma, or a tumor originating in a major or minor salivary gland. However, as the clear bordered cystic nature of the mass was well described from the MRI, together with the past treatment references from other local clinics, in this case, we diagnosed rarer expanded plunging ranula.

The patient had already received fenestration of the cyst wall twice in other hospital, so her parents strongly request to treat more definitely this time. We explained to her parents that the benefit and the drawback of each surgical procedures including the fenestration, local injection of the OK-432, cystectomy and then more thoughtfully and carefully, we, then, decided the treatment plan

that the extirpation of the sublingual gland together with the fenestration of the cyst pursuing the parapharyngeal space to get healing completely.

The surgery of extirpation of the sublingual gland together with the fenestration of the cyst into the parapharyngeal space were performed under general anesthesia using an intraoral approach. Following the fenestration of the cyst wall, the cyst cavity was then communicated with careful attention from the submandibular to parapharyngeal spaces using mosquito forceps. Front end of the length of 5 cm penrose drain was then reached to the infratemporal fossa through the parapharyngeal space and that was placed in the oral floor (Fig. 3). Total operation time was 60 min without striking blood loss, and her ingestion was started on the same day of surgery. No lingual nerve problems ever or surgical site infection occurred postoperatively with uneventful clinical course. The patient was discharged well on the fourth postoperative day.

The histopathologic diagnosis was plunging ranula (Fig. 4). The Penrose drain was removed 2 weeks after the surgery. An MRI performed 6 months postoperatively showed that the lesion had disappeared completely (Fig. 5) and 12 months postoperatively further confirmed no recurrence at the both intraoral and extraoral sites (Fig. 6). One and half years have passed postoperatively, no signs of recurrence was seen after the regular follow-up so far.

3. Discussion

A plunging ranula is generally known to develop when extravasation of mucus extends through or around the mylohyoid muscle and presents with swelling of the oral floor. Such lesions are usually of the sublingual or submandibular type. At the level of the mylohyoid muscle, the parapharyngeal space lacks any facial boundary that separates it from the sublingual and submandibular space. Thus, there is open communication among these three spaces, allowing lesions either the sublingual or submandibular space to enter the parapharyngeal space [3]. The expansion of a ranula into the parapharyngeal space is rare, with only 32 cases identified in the literature [2,4–8]. Expansion further into the infratemporal fossa is even more rare, and there are no English published case reports describing this presentation so far.

In our case, a local doctor had performed incision and fenestration of the cyst wall on two occasions before referring the patient to our department. Given that a plunging ranula usually expands to the submandibular space over the mylohyoid muscle

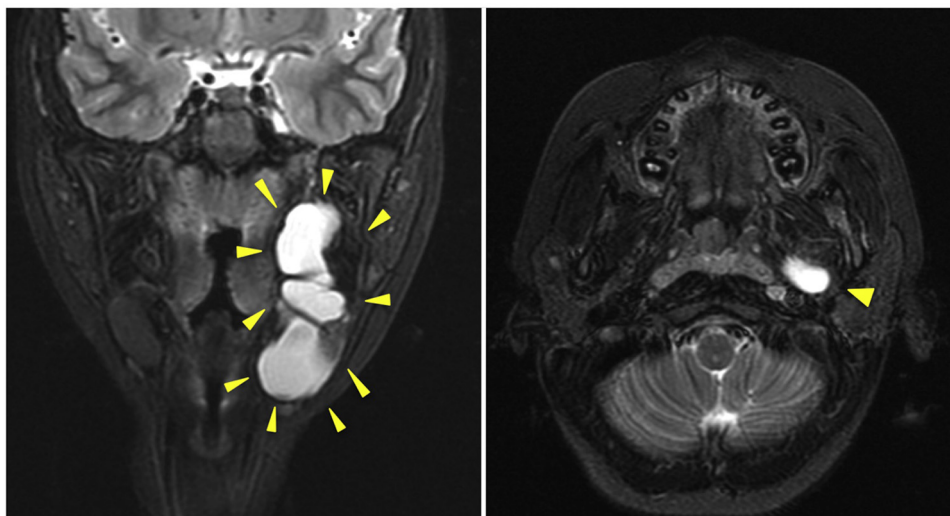


Fig. 2. Preoperative magnetic resonance imaging. A cystic lesion of high density seen on T2 weighted imaging, located mainly in the left submandibular space but expanding to the parapharyngeal space and infratemporal fossa (arrow heads).

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