Bilateral plunging ranula: two case reports and a review of the literature

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Abstract

Aim Presentation of two bilateral plunging ranula cases and then review of the plunging ranula literature to understand current concepts on aetiology, imaging for diagnosis and management.

Method A literature review using PubMed (92 papers) and Google Scholar (18 papers) has revealed only 18 cases of bilateral plunging ranulas ever published and with the addition of the two cases presented this makes a total of 20.

Results These cases are reviewed and information related to aetiology and management is presented. The cause appears to be a combination of mylohyoid dehiscence, racial predisposition and previous trauma to the mouth/face or previous oral surgery.

Conclusion Plunging ranula are a rare cause of bilateral and unilateral neck swellings but more common in Māori, Polynesian and Asian people. Their cause is multifactorial and ultrasound scan (USS) is the current investigation of choice. Management relies on excision of the sublingual gland with the cystic contents via a trans-oral approach.

The term ranula is derived from the Latin word “rana” meaning little frog and is descriptive of the blue translucent swelling in the floor of the mouth, which is said to resemble the under belly of a frog¹. A simple ranula is the result of extravasation (leakage) of saliva from any of the 20 ducts that arise from the sublingual gland and empty into the floor of the mouth or into the anterior portion of the submandibular duct².

Ranulas are characteristically large (3–6 cm) and form a blue, tense vesicle in the floor of the mouth. Some ranulas will attain sufficient fluid pressure to herniate through the mylohyoid muscle into the submandibular space within the neck and are termed plunging or diving ranulas. A plunging ranula usually presents as a painless fluctuant lateral neck swelling which does not change with swallowing or eating.¹ It is most commonly centred within the submandibular space and averages 4–10 cm in size.

Bilateral plunging ranula are very rare with only 18 cases reported in the literature. A broad differential diagnosis exists for unilateral neck swellings and this also applies to bilateral swellings. Other causes of submandibular swelling that need to be considered in both the community and hospital settings include submandibular space abscess/collection from an odontogenic source, metastatic disease from cancers of the lip, skin of the face and oral cavity and tumours of the submandibular gland (50% of which are malignant). Other cystic lesions that present within this position in the neck
include dermoid cyst, cystic hygroma (lymphangioma) or haemangioma. Additionally adolescents may develop acute inflammatory lymphadenopathy from bacterial and viral infections such as glandular fever.

It has been widely suggested that the cause of a plunging ranulas is due to a congenital predisposition involving dehiscence of the mylohyoid muscle, allowing the sublingual gland to herniate into the cervical tissues, and racial predisposition.\textsuperscript{1,3,4} There is also a possible correlation with trauma to the floor of mouth and sublingual duct rupture\textsuperscript{5} or previous oral surgery.\textsuperscript{3}

A unilateral plunging ranula is a rare entity with the chances of it occurring bilaterally even less likely but should be considered by general practitioners, emergency medicine, ear nose throat and oral/maxillofacial specialists when presented with neck lumps. We describe two cases with one having had previous trauma to the mandible and both having radiological evidence of bilateral dehiscence of the mylohyoid muscle and herniation of the sublingual gland.

**Case 1**

A 20-year-old fit and well Māori man, presented with a left-sided submandibular swelling. He had sustained trauma to the left side of his face 4 days prior to presentation. Initially there was minimal swelling but was followed by a rapid left submandibular pain and swelling over the next day with extension down the lateral neck.

The swelling was firm and diffuse, extending from the left inferior border of mandible to thyroid cartilage inferiorly. Wharton’s duct was patent and he had no cervical lymphadenopathy. Bloods and a panoramic film were unremarkable.

Computed tomography (CT) neck demonstrated a multiloculated rim enhancing fluid collection around the left angle of the mandible within the submandibular space (Figure 1). The possibility of an anterior mylohyoid defect on the left side was raised. It was also noted that there was an additional plunging ranula on the right side with herniation of sublingual gland posterior to this defect on the right side. The mylohyoid defect was within the anterior two-thirds of the muscle.

**Figure 1. Axial CT showing fluid collections within bilateral submandibular spaces**

![Axial CT showing fluid collections within bilateral submandibular spaces](image-url)
This mylohyoid defect was better demonstrated on ultrasound scan (USS) through which a small amount of sublingual gland was seen to herniate. See Figure 2.

**Figure 2. Ultrasound scan of the neck showing the mylohyoid (MH) dehiscence**

At this stage the decision was made to address the symptomatic left plunging ranula with aspiration, incision and drainage of the collection and then left sublingual gland excision via a trans-oral approach with removal of the cyst contents. The aspirate was positive for amylase. Histology showed focal mucous extravasation and patchy chronic inflammation of the gland.

Unfortunately 6 weeks later, he developed a sudden right-sided neck swelling. The right sublingual gland was then excised in a similar fashion to the left with access via a trans oral approach. A small part of the herniated sublingual gland was lifted from the neck below the mylohyoid muscle dehiscence. The histology report was similar to the left gland with mucous extravasation and chronic inflammation. He has remained asymptomatic in subsequent clinic reviews.

**Case 2**

A 21-year-old Māori man presented with a 3-day history of a painless, enlarging, fluctuant left neck swelling which he initially described starting after having a sore throat and it enlarging over the course of a day. He was otherwise well and denied any dental pain or trauma.
The left submandibular swelling was non-tender and extended down the lateral neck, measuring 10 cm in diameter. He had no intraoral signs of odontogenic infection. His floor of mouth was soft and he had neither trismus nor cervical lymphadenopathy. CT neck confirmed the presence of bilateral plunging ranulas, with the symptomatic left plunging ranula larger than the right.

He underwent aspiration, incision and drainage of the left submandibular space. The drained fluid was clear and mucoid in nature and was free of pus. Biochemical analysis revealed the "straw-coloured" fluid being mucous and saliva.

The patient made an uneventful recovery but unfortunately failed his outpatient review and has been lost to follow-up before the definitive excision of the gland was able to be offered.

Discussion and review of the literature

A literature review using PubMed (92 papers) and Google Scholar (18 papers) has revealed only 18 cases of bilateral plunging ranulas ever published and with the addition of the two cases presented this makes a total of 20.

A total of nine of the 20 cases have been reported from the Auckland region and has led to the suggestion that there is a congenital predisposition in people of Polynesian and Māori descent to have a defect in the mylohyoid muscle, which forms a muscular diaphragm in the floor of the mouth, allowing ectopic sublingual gland tissue to escape below the muscle.4

All but one of the cases are from either New Zealand or the Asia-Pacific region. This aberration in the mylohyoid muscle has been described before, reported by Engel (1987)6 and other papers have documented this observation.1,3-5,7,15

If people of Polynesian and Māori descent have a genetic predisposition to having a unilateral dehiscence in their mylohyoid muscle it is quite reasonable to assume that this sometimes occurs bilaterally, which may predispose some individuals to having bilateral plunging ranulas.

Four bilateral cases are reported in a recent large review of 77 patients by Morton et al.4 Their surgical findings support the theory of a dehiscence in the mylohyoid muscle in 67 of 69 operated cases. They also comment there is a strong predilection to Māori and Pacific Island people and that there may be a genetic component to this condition with Māori and Pacific Island people making up 82% of the cases.

Morton et al published a case series of 20 patients over a 9-year period in the Auckland region and 100% were Māori or Pacific Islanders.4 Along with other large studies most have a common racial origin as demonstrated by over 80% of all cases being of Asian descent.1,4,8,9

Further weight to this hypothesis comes from Mahadevan and Vasan3. They published a case series of 21 paediatric patients from the Auckland region over a 5-year period with 20 of them being Māori and Pacific Islanders. There were three bilateral plunging ranulas in a 6, 9 and 15 year old respectively.

Previous blunt trauma to the face/neck and previous head/neck surgery were identified as further risk factors for plunging ranula development with 9 of 21 patients having had surgical procedures such as submandibular gland excision prior to
developing the plunging ranula.\textsuperscript{3} Tail et al also reported six patients developing intraoral ranulas after submandibular duct transposition surgery.\textsuperscript{10} There are a further 11 bilateral plunging ranula cases in the literature.\textsuperscript{8,9,11-14} All but one are from Asian countries, although a British case appears to have been the first, described by Barnard in 1991.\textsuperscript{11} Trauma and previous oral surgery are often mentioned in these case reports as part of the history.

The diagnosis of plunging ranula can be difficult and different techniques have been employed over the years, ranging from aspiration and examination of the cyst fluid for salivary amylase and protein content\textsuperscript{11} or fine needle aspiration and cytology\textsuperscript{3} to imaging with MRI, CT and now ultrasound scan (USS) becoming the modality of choice.\textsuperscript{15}

Jain et al investigated 33 cases of plunging ranulas over 4 years with high resolution USS.\textsuperscript{15} The potential benefits of this investigation include measuring the extent and dimensions of the plunging ranulas, confirming the cystic nature and the status of the mylohyoid muscle (100\% of cases had a defect) and evaluating the sublingual gland for rupture and herniation. Patients in the study had a median age of 20 years old and USS would avoid radiation exposure to these younger patients.

Various surgical approaches to excising the plunging ranula and/or the sublingual gland have been described in the papers reviewed, through either a trans-oral or cervical approach. Some have recommended excision of the pseudo cyst also\textsuperscript{3} or inducing sufficient fibrosis to seal the mucous leak.\textsuperscript{5}

The optimum surgical treatment has been proposed by Morton and colleagues from their large South Auckland experience and relies on complete trans-oral excision of the sublingual gland and evacuation of cervical cystic contents.\textsuperscript{16} A trans-oral approach to the gland was the preferred method in the majority of the cases reviewed and is the technique of choice in our department.

**Competing interest:** None known.

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