

A retrospective study of ranula in two centres in Malaysia

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Abstract

Introduction Ranula is a mucous extravasation cyst which occurs as a result of trauma or obstruction of the sublingual or minor salivary gland or the duct itself.

Patients and results 14 patients were seen at Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur and Hospital Tunku Ampuan Afzan, Kuantan, Malaysia between 2000 to 2006. There were six cases of intra-oral ranula and eight plunging ranula.

Twelve patients underwent surgical intervention while two refused surgery. Four patients (33.3%) from those who had surgical intervention returned with recurrence; two (16.7%) had marsupialization and the other two (16.7%) had excision of the pseudocyst intraorally.

Conclusion Excision and marsupialization remain as the treatment of choice in our centres.

Keywords Ranula · Intraoral · Plunging · Malaysia

Introduction

Ranula is a pseudocyst caused by mucous extravasation into the surrounding soft tissue in the floor of the mouth as a result of trauma or obstruction to the sublingual or minor salivary glands or the duct itself. Although it is commonly seen intraorally, it can be located extraorally as a submental or submandibular swelling and known as plunging ranula. A ranula is relatively painless or asymptomatic with little or no associated morbidity or mortality but if the size is large, it may affect swallowing, speech, mastication or breathing.

Surgical excision of the sublingual gland together with or without the cyst has been regarded as the best option in view of the least recurrence rate [1]. However, marsupialization which is less invasive is now becoming widely accepted and recommended particularly in children [2–4].

This study reviews our experience in the treatment of ranula and its outcome.

Patients and methods

We retrospectively reviewed records of patients who were diagnosed with ranula at the Universiti Kebangsaan Malaysia Medical Centre, Kuala Lumpur, Malaysia and Tengku Ampuan Afzan Hospital, Kuantan, Pahang, Malaysia.

Data extracted from the records included:

1. Demographic data i.e., age, gender, race
2. Age of presentation and age when the swelling was first noticed
3. Site/s of swelling: whether the swelling was confined intraorally only or involved intra and extra-oral
4. Associated systemic diseases
5. Symptoms associated with ranula
6. Imaging
7. Surgical methods: excision or marsupialization
8. Outcome of the treatment

The period of follow-up was from 6 months to 2 years. Outcome of treatment in particular involving recurrence was evaluated. A recurrence is described when

a similar saliva-contained swelling recurs in the previously operated area.

Complications such as bleeding, nerve injury or Wharton's duct injury were also assessed.

Results

14 patients, six males and eight females between the age of second day of life to 44 years old (mean age 15.3 years old) were seen from June 2000 to February 2006. The patients were twelve Malays and two Chinese.

There were six cases of intra-oral and eight plunging ranula. Two cases presented with difficulty in breathing and swallowing, one of which being associated with secondary infection and one case with difficulty in feeding. Thirteen patients were medically fit. Only one patient was diagnosed with hepatoblastoma and malignant hypercalcemia alongside with ranula.

In relation to investigations and imaging, two patients had Magnetic

Resonance Imaging (MRI), four patients had Computer Tomographic (CT) scan, one had Orthopantomogram (OPG), one had Postero-Anterior (PA) mandible and lateral skull radiograph and another patient had a sialogram as sialolith was thought to be a contributory factor (Figs. 1 and 2).

Five patients did not have any form of investigation. Fine Needle Aspiration (FNA) was carried out on one patient.

Five patients had marsupialization, four had excision of the pseudocyst intra-orally, three had excision of the sublingual gland and two refused surgery.

Outcome of treatment especially involving recurrence was evaluated.

Four patients (33.3%) from those who had surgical intervention returned with recurrence; two (16.7%) had marsupialization and the other two (16.7%) had excision of the pseudocyst intraorally.

Discussion

A ranula can be classified into two clinical types according to the sites of primary swelling. An oral ranula is located in the floor of the mouth. Clinically, it is dome-shaped with bluish discoloration and may fill the mouth and raise the tongue. It is typically painless and does not change in size in response to chewing, eating or swallowing. A plunging ranula is located near the upper airway and extends into the floor of the mouth. From the submandibular space, it can extend into the submental region, the contralateral side of the neck, the nasopharyngeal area and up to the skull base, the retropharyngeal and into the upper mediastinum [5,6].

A congenital ranula may develop as a result of failure in the development of the salivary duct. An acquired aetiology is usually related to trauma to the sublingual gland or the duct and scar formation following this may cause obstruction.

Plunging ranula may form because of dehiscence in the mylohyoid muscle via an ectopic gland or via an anatomic association with the submandibular gland allowing the pseudocyst to pass around the posterior border of the mylohyoid.

In relation to demographic factors, no racial predilection has been reported but from our study, the Malays showed the most preponderance (85.7%) and Chinese, a 14.3%. Our results showed male to female ratio of 1:1.3.

Ranula is not a common pathology in the oral cavity. It occurs from 1% to 10% and has a prevalence of 0.2 cases per 1000 persons [7].



Fig. 1 Two-day old infant with ranula

In this study, the occurrence of plunging ranula (57.1%) was more common than oral ranula (42.9%). This was in contrast with a study from Zimbabwe but in agreement with the New Zealand's study [8,9].

Majority (42.9%) of the patients in this study were younger than 10 years old with the youngest at day 2 of life, 35.7% in the 11 to 20 years old group, 7.1% in the 20 to 30 years old and 14.3% in the above 35 years old group. Chidzonga et al. also showed that the peak prevalence in their study was in the 0 to 10 year old age group, while Zhao et al. showed an older cohort [8,10].

There were various methods of investigations of a ranula reported in the literature ranging from clinical observation alone to FNA and imaging. Imaging modalities include ultrasound, CT scan and MRI [9,11]. The MRI is the imaging of choice to delineate the extent of lesion and its association with the normal structures. FNA in ranula usually revealed high protein and amylase concentrations. Increased levels of matrix metalloproteins, tumour necrosis factor- α [12], type IV collagenase, plasminogen activators and proteolytic enzymes were shown to be responsible for the invasive character of the extravasated mucous [13].

The choice of investigation is entirely depending on the surgeons' preferences.

Mahadevan and Vasan preferred FNA in all their patients and no radiological investigations was performed in their case series [9], and this was in agreement with Zhi et al. [14]. On the other hand, a study from Italy revealed that the choice of imaging was ultrasound tomography in all their patients [15]. Our study showed the use of different modalities in investigation and this was in agreement with a study from Korea [16].

A ranula can be presented for years without revealing any other symptoms

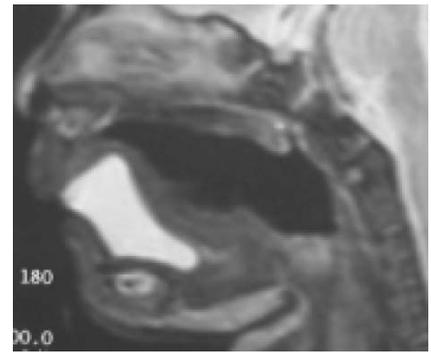


Fig. 2 MRI showing extension of the cyst in the oral cavity

except for swelling. Therefore, removal can be delayed except in cases where the airway or the upper aerodigestive tract is obstructed. In our study, two of the patients showed difficulty in breathing and feeding and one case with difficulty in swallowing.

In relation to systemic disease or condition, there was no associated illness in all patients except in one child who was concurrently diagnosed with hepatoblastoma, therefore we could not correlate ranula to any of systemic diseases or congenital malformations. However, case series from Zimbabwe revealed possible association of ranula with Human Immunodeficiency Virus (HIV) [8,17].

To date, there are many interventions to treat ranula in the form of surgical or non-surgical. Excision of the sublingual gland with or without the cyst is the treatment of choice in view of the least risk of recurrence (from 0% to 1%) [8,15,18].

Marsupialization on the other hand, is a procedure in which the cyst is unroofed and the lining is sutured to the surrounding mucosa to create a patent communication with the oral cavity. This technique is less invasive compared to excision however, is associated with high recurrence rate, approximately 60% to 90% [4]. To overcome this, Baumash advocated marsupialization and packing of the cyst and this reduced the recurrent rate from 10% to 12% [3,4]. The gauze pack is placed into the depths of the cavity and its maintenance in the position will provide an optimal pressure. This was supported by Mortellaro et al. where an iodoformic gauze pack was placed in the cavity and showed that the recurrence was observed to be halved (12%) compared to recurrence from the conventional technique [15].

In our centres, surgery was carried out using various techniques i.e. from excision of the ranula intraorally to marsupialization or excision of the sublingual gland and a combination of any of these. The reasons

Table

No.	Sex	Age at presentation	Race	Noticed ranula since	Intra-oral vs plunging	Other illness / symptoms	Imaging	Treatment	Outcome after treatment
1.	Female	7-years-old	Malay	7-years-old	Plunging	NOI	Nil	Excision of sublingual gland	No recurrence
2.	Female	17-years-old	Malay	15-years-old	Plunging with cellulitis	Difficulty in breathing and swallowing	PA mandible and lat skull	Marsupialization	Recurrence after a year but resolved after rupture
3.	Female	9-years-old	Malay	8-years-old	Intraoral	NOI	Sialogram	Marsupialization	Recurrence after seven months and re-marsupialized then no recurrence
4.	Female	24-years-old	Chinese	Not recorded	Intraoral	NOI	OPG	Refused surgery	Swelling disappeared after ruptured
5.	Female	44-years-old	Chinese	8-years-old	Plunging	NOI	MRI+CT scan	Refused surgery	Swelling still present
6.	Male	Day 2 of life	Malay	Birth	Plunging	Difficulty in breathing and swallowing	MRI	Marsupialization	No recurrence
7.	Male	9-months-old	Malay	Birth	Intraoral	Difficulty in feeding	Nil	Marsupialization	No recurrence
8.	Male	4-years-old	Malay	4-years-old	Intraoral	NOI	Nil	Excision of sublingual gland	No recurrence
9.	Female	1-year-old	Malay	Birth	Intraoral	Hepatoblastoma and malignant hypercalcemia	Nil	Marsupialization	No recurrence
10.	Male	17-years-old	Malay	16-years-old	Plunging	NOI	CT scan	Excision of pseudocyst	Recurrence after a year
11.	Male	18-years-old	Malay	Not recorded	Intraoral (infected)	NOI	CT scan	Excision of pseudocyst	No recurrence
12.	Male	17-years-old	Malay	17-years-old	Plunging	NOI	CT scan	Excision of sublingual gland	No recurrence
13.	Female	19-years-old	Malay	17-years-old	Plunging	NOI	FNA	Excision of pseudocyst	No recurrence
14.	Female	37-years-old	Malay	Childhood	Plunging	NOI	Nil	Excision of pseudocyst	Recurrence after six months

Note:

- PA – Postero-Anterior
- Lat – Lateral
- OPG – Orthopantomogram
- MRI – Magnetic Resonance Imaging
- CT – Computed Tomogram
- FNA – Fine Needle Aspiration
- NOI – No Other Illness
- Nil – No Imaging done

for various surgical techniques were based on the age of the patient, the size of the pseudocyst and surgeons' level of experience. In our study, marsupialization was normally performed in patients aged one year old or below as the access to the floor of the mouth in this group of patients is very limited. In addition, marsupialization allows removal of a smaller amount of tissue with better defined interfaces and less likelihood of injury to the nerve and Wharton duct.

On the other hand, excision was carried out in larger ranula and plunging ranula except in a case which involved a 17-year-old boy with an infected plunging ranula. His clinical manifestations included difficulty in breathing and swallowing which could be related to secondary infection. Marsupialization was carried out for decompression. Although it recurred a year later, the condition resolved after a spontaneous rupture.

On the whole, marsupialization is still very popular as it carries low risk of complications compared to excision of the sublingual gland. In this study, the total recurrence rate was 33.4%; 16.7% from marsupialization and 16.7% from excision of the pseudocyst. No recurrence was observed in patients who had excision of the sublingual gland. In the literature, there were a few complications observed with sublingual gland removal namely, injury to Wharton's duct (2%), bleeding (1% to 2%), infection (1% to 2%) or paraesthesia of the lingual nerve (2% to 12%) [1]. In this series, we encountered a case of postoperative haematoma which completely resolved after a month.

Spontaneous regression was observed in two patients, one with recurrence and another who refused surgery. This could be due to trauma during mastication that causes rupture of the cyst lining and this depressurizes the cyst. A patent communication between the cyst and the oral cavity could prevent the cyst fluid from accumulating within the cavity.

Other surgical methods involved carbon dioxide laser and fenestration and continuous pressure [19,20].

Non-surgical treatment of a ranula includes intracystic injection of OK-432 (Picibanil; Chugai Pharma Co, Tokyo, Japan) [11], an inactivated form of streptococcus pyogenes, botulinum toxin type A [21] and oral administration of Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations

[22]. The authors of these articles claimed that all these alternative modalities showed a high success rate of more than 80%.

Conclusion

Although many interventions have been proposed with reasonable success rate, surgery still remains the mainstay of treatment. Excision of ranula with the sublingual gland is the preferred method to avoid recurrence but in circumstances where the access is difficult especially in young children, marsupialization with packing is advocated. Both excision and marsupialization remains as the treatment of choice in our centres.

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