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Antique Method of Management of Sublingual Ranula in Children under General Anaesthesia

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Abstract:
Background: Ranula is a mucocele which occurs on the floor of the mouth caused by mucus extravasation of the sublingual gland.The term ranula is derived from a latin word 'rana' meaningfrog.The most common clinical presentation is swelling under the tongue. Due to involvement of the floor of the mouth, ranulas always poses a challenging situation surgically, **Case Presentation:** The objective of this article is to report a case of a ranula in five-year-old female child complaining of a painless reddish swelling below the tongue on the left side for the past one week. The patient undergone surgical excision under general anaesthesia, **Conclusion:**Management of ranula by surgical excision had no recurrence at 6 months and it proved to be an effective treatment method.

Keywords: Floor of Mouth, Salivary gland diseases, Ranula, Sublingual Gland

Introduction:

The ranula is a cystic mass which is filled with mucin which originates from extravasation of the sublingual gland (Mahadevan et al., 2006).Mucin pooling from ruptured salivary gland ducts or acini causes Ranulas (Choi MG, 2019).The intraoral swelling looks similar to a frog's translucent underbelly(Mustafa et al., 2013).It develops due to trauma or obstruction of the excretory duct of the salivary gland situated in the submandibular or sublingual space (Sheikhi et al., 2011). It usually presents in the floor of the mouth as a well circumscribed, soft, bluish swelling covered by a thin layer of epithelium. Ranulas are commonly huge (>2cm) and look like a dome shape vesiculous lesion (Chauhan et al., 2014). It is classified as simple (intraoral) ranula and plunging (cervical) type of ranula. A simple ranula represents a localised collection of mucous within the floor of the mouth,whereas in case of plunging ranula, the mucous collects in the submandibular and submental spaces of the neck with or without intraoral

involvement (Lloyd et al., 1995).Based on their location, it can be classified as Sublingual, Sublingual-submandibular and Submandibular type of ranula. Simple ranula involves the sublingual gland (Yang et al., 2014). Whereas in plunging ranula, there is involvement of sublingual and submandibular gland. The fluid pressure of the mucus dissecting to the submandibular gland through the mylohyoid muscle (Langlois et al., 1992).The prevalence of ranula accounts for about 6% of all oral sialocysts. Only 1 in 10 ranulas are true retention cysts. In children and young adults, ranula occurs commonly. The peak age range of ranula is in the second decade of life, however plunging ranulas are seen most commonly in third decade of life (Zhao YF et al., 2004).The prevalence of paediatric ranula was 1.15:1 (F:M) with a slight predilection towards the female population(Langlois et al., 1992).Investigations prior to surgery include ultrasonography (USG), computed tomography (CT) and Magnetic Resonance Imaging (MRI). After surgery, sialochemistry and histopathological analysis is recommended.Various treatment modalities for ranula have been suggested in the literature like simple aspiration or drainage of ranula, marsupialisation, ranula excision, sublingual gland excision with ranula evacuation, sublingual gland plus ranula excision, submandibular and sublingual gland excision plus ranula excision, CO2 Laser, radiation Therapy, sclerotherapy and oral administration of Nickel Gluconate-Mercurius Heel-Potentised Swine Organ Preparations D10/D30/D200, and each treatment has shown a diverse success rate (Nguyen et al., 2017).Many newer conservative methods are also promising and can be used initially for treatment of ranula in selected cases. However, surgical excision is the most reliable method according to the literature.Therefore, this case report describes the cases of simple ranula and and its management under General anaesthesia.

Case presentation:

A 5-year-old female child presented to our OPD, Department of pedodontics with a chief complaint of a painless reddishswelling below the tongue on the left side for the past one week. The parent gives a history of similar swelling in the same area six months back, which gradually decreased in size without any treatment. There was no history of traumatic injury, infection or any other triggering factors. Paresthesia was not associated with the swelling. On clinical examination, a swelling measuring 2 x 3 cm^2 in the ventral surface of the tongue with extension from posterior aspect of mandibular primary second molar up to 1cm behind the corner of the lip in the lefts sublingual region of the floor of the mouth (Figure 1). The swelling was non tender,soft and non-compressible on palpation. Swelling was not associated with any ulcer, pus discharge. There were no signs of any cervical extensionThe swelling was provisionally diagnosed as a ranula. A customary bloodinvestigations were done;the lesion was exposed using an intra oral approach.

After routine blood investigation, intraoral approach for surgical excision of ranula was planned under general anesthesia. Informed consent was obtained from the parent. After intubation, a topical anesthetic gel (benzocaine 20%) was applied over the surface of lesion and deroofing of the cyst was done and there was drainage of mucin mixed with blood, which confirmed our diagnosis of ranula (Figure 2). Discharge was removed followed by excision of ranula (Figure 3) and 3-0 Black braided sutures were placed to close the mucosal defect (Figure 4). Patient was discharged after one day since the post-operative period was uneventful. The child and the parent received post-operative instructions on diet, toothbrushing and advised application of 0.5% chlorhexidine gel to the sutured site for 7 days post-operatively to prevent secondary infection.

The excised specimen was sent to histopathological examination. It revealed the presence ofmixed mucous and serous glands with mucin surrounded by chronic inflammatory infiltrate, without evidence of malignancy.

The patient was reviewed after a week and it was seen that patient was asymptomatic, the lesion partially regressed and the sutures were removed. Again the review was done after 15 days and after one month. On one month follow up, there was complete regression of the lesion and at 6 months follow up, no clinical signs of recurrence were seen.

Figure 1: Preoperative Intraoral photograph showing swelling on the left side of the ventral surface of the tongue

Figure 2: Surgical excision of ranula

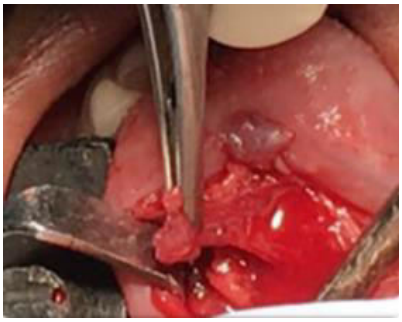
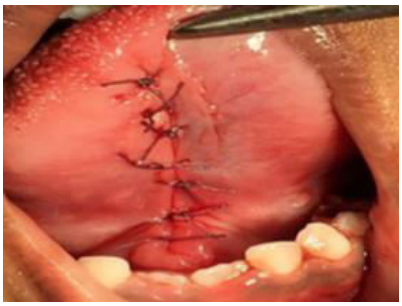


Figure 3: Surgical site after excision of the ranula

Figure 4: Post-operative photograph after suturing of the defect



Discussion:

Since the time of Hippocrates and Celsius, ranula has been known and it is used to describe a pseudocyst(Verma et al.,2013).Initiation of ranula formation occurs by mucus accumulation in the surrounding connective tissue without an epithelial lining describing it as a pseudocyst(Quick et al., 1977).

Biochemical investigations reveal that saliva reveals high protein and amylase content along with secretions from the acini of the sublingual gland. Due to its high protein content in the secretion it produces a very intense inflammatory reaction and mediate pseudocyst formation(Morton et Al., 1995).Ranula occurs due to two processes. One is the formation of an epithelial-lined retention cyst due to partial obstruction of a sublingual duct. It is rare and occurs only in less than 10% of all ranula. Secondarily, it can occur due to trauma(Godhi et al., 2013).

If a duct is obstructed, there is build-up of secretory back-pressure leading to salivary duct rupture with mucus being forced into the surrounding tissues and also trauma can directly damage the duct or acini, leading to mucus extravasation and forms a pseudocyst(Godhi et al., 2013).Oral simple ranulas present with intraoral swelling only, plunging ranula presents with cervical swelling without swelling of the floor of the mouth. A mixed ranula involves both intraoral and cervical swelling(Zhao et al., 2004).The diagnosis of ranula is based on various investigative methods. The various differential diagnosis includes inflammatory and neoplastic lesions of the sublingual and submandibular glands/lymph nodes, granulomatous, adipose tissue diseases, cystic hygroma, branchial or

thyroglossal duct cysts, laryngocele, dermoid cysts, and epidermoid cysts(Lee et al., 2015).Lee DH et al. (2015) conducted a retrospective cohort study and reported that surgical excision of ranula along the sublingual gland by intraoral approach is the optimum treatment of simple ranula.Yang Y and Hong K(2014)performed a prospective cohort study and found that removal of ranula along with the associated salivary gland is the safe and effective method.Clyburn et al, (2009) published a case report where they have treated the ranula by resecting the submandibular gland if ranula extends into the neck and pharyngeal spaces using transcervical approach.Yuka et al. (2005)did a retrospective cohort study and suggested that marsupialization is the effective method for treatment of ranula to prevent recurrence and also reported that it should be combined with sublingual gland. Hidaka et al. (2003) published a case series where they showed that surgical extension of the cyst along with the sublingual gland via intraoral approach is the best approach for the management of plunging ranula.Bozena et al. (2015)reported that surgical excision of the cyst along with the sublingual gland was effective with no recurrence at 10 years follow up. Crysdale et al. (1988) reported that the success rate was 100% when the ranula was excised along with the underlying sublingual gland if the ranulas are greater than 1 cm. If the ranula recurs after marsupialization, the involved sublingual gland must be removed.

There are multiple approaches for treatmentof ranula. It can be treated by surgical excision of the cyst with or without involvement of the sublingual gland, cryosurgery, marsupialization, cryosurgery, radiotherapy and CO2 laser excision. Before planning with the surgical method, spontaneous resolution may be another option for paediatric patients. It is recommended to wait and watch for the lesion to dissolve till 6 months(Lloyd et al., 1995). If it is not dissolving then, surgical treatment is indicated. There is no consensus in the literature regarding the recurrence rate. Few authors have claimed that conservative treatments like marsupialization havehighest recurrence rate(Nguyen et al., 2017).

There are few complications which may arise following transcervical surgical approaches than the intraoral approach such as scar formation and damage to the mandibular nerve. The rate of recurrence was also found to be lower in the intraoral approach(Lloyd et al., 1995). Therefore, in our case we have selected intraoral approach to excise the lesion. Similarly, study done by Zhao et al. (2004) did a research to find out the recurrence rates of 580 treated ranulas which was managed using different surgical methods. They showed that there was no difference in the recurrence rates based on the type of ranula but there was difference in the surgical method. After marsupialization it was 66.7%, and 57.69 % after surgical excision of ranula and 1.2 % after excision of the sublingual gland with or without the ranula(Zhao et al., 2004).In this case, there were no

signs of recurrence at 6 months' follow-up period since all the mucous were completely extracted from the surgical site.

Conclusion:

The salivary gland disorders depend on accurate diagnosis and effective treatment. Among various treatment modalities,Surgical removal of ranula along with the underlying sublingual gland is the most preferred, reliable treatment to prevent recurrence.

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