2 CASE REPORTS OF PEDIATRIC RANULA WITH A SYSTEMATIC METANALYSIS

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ABSTRACT
A ranula a mucous filled cavity by definition; commonly seen in the floor of the mouth within the sublingual gland. Based on clinical features they are of various types. The incidence of ranula in pediatric group is hardly evaluated in depth. The imaging modalities of choice include ultrasound, CT and MRI. Surgical management is the commonly opted treatment though other modalities are available; and can be attributed to the increased chances of recurrence. Through this paper we highlight two cases on ranula in pediatric patients reported to our department within a period of one week; with a review of literature emphasizing the use of the advanced imaging diagnostic methods and updated medical line of treatment.

KEYWORDS: Ranula, Sublingual gland, ultrasound for ranula.

INTRODUCTION
Ranula’s present as cystic dilatations in the floor of the mouth that occurs due to rupture or damage of ducts of the sublingual glands leading to mucus extravasation or dilatation of the gland's duct¹¹. It can be of extravasation or retention type; the latter is more common. Literature quotes that they can be even congenital; but congenital ranula in newborn infants is reported in rarity and thus there is a marked paucity of the literature on the cited subject¹².

CASE REPORT 1
A 5 year old girl reported to Department of Oral Medicine & Radiology with a complaint of a swelling in the floor of the mouth, since 2 days, that was nontender and increasing in size.
History further revealed that the growth was gradual on onset and progressive in size and was painless throughout its course. History of trauma induced by tooth brush 3 days back was elicited from the patient on careful history. On clinical examination a presence of a single well circumscribed bluish dome shaped swelling on the right floor of the mouth, measuring approximately 2.5 X 2cm in size. The swelling is smooth surfaced, with no signs of ulceration or discharge. Overlying mucosa was stretched and translucent. The mucosa surrounding the swelling appears to be normal. The swelling was lifting the tongue on the right side. The swelling was non-pulsatile (Fig - 1,2).

On palpation the inspectory findings of site, size, shape and extent were confirmed. The swelling was non-tender, soft in consistency, fluctuant, compressible and reducible on palpation. Diascopy test was negative. On aspiration of the swelling it yelled a viscous yellowish tinged fluid. After correlating the history and clinical findings, case was provisionally diagnosed as “ranula”.

The patient was subjected for radiographic examination. Occlusal radiographs: revealed no relavent radiographic features (Fig - 3). Ultrasound reveals An illdefined cystic lesion of size 1.1 X 0.9 X 0.4, volume 0.2-0.3cc is seen in the right sublingual region. Suggestive of ranula (Fig - 4).

After all clinical and preoperative evaluations excision of ranula along with the sublingual gland was done under general anaesthesia. The excised sample was sent for histopathological examination, which revealed predominance of histocytes in the cystic space and on the pseudocystic fibrous connective tissue wall and further the central cystic space and the walls composed of loose and vascularized connective tissue with the sublingual gland suggestive of mucous retention cyst (Fig - 5). Child was under follow up for the past 6 months and there was no recurrence.
Case Report 2
A four and half year old girl reported to Department of Oral Medicine & Radiology with a complaint of a swelling under her tongue on the left side since 10 days. History of present illness revealed that the swelling is of the same size since it was noticed. There was difficulty in mastication.

On clinical examination a presence of a solitary swelling evident in relation to the floor of the mouth on the left side of the lingual frenum measuring approximately 4 x 2.5 cm in size. It is dome shaped, well defined, has a smooth surface and the mucosa covering the swelling appears to be stretched. A bluish hue is present on the swelling. The swelling appears to be lobulated and tongue appears raised. Mucosa surrounding the swelling appears normal (Fig - 6,7).
On palpation inspectory findings of site, size, shape and extent were confirmed. The swelling is soft consistency and is fluctuant. It has well defined borders and is compressible. No sign of tenderness on palpation. On aspiration of the swelling it yielded a viscous yellowish tinged fluid. After correlating the history and clinical findings, case was provisionally diagnosed as “ranula”.

The patient was subjected for radiographic examination. Occlusal radiograph (Fig - 8): revealed no relavent radiographic features. Ultrasound reveals a mildly bulky and heterogenous submandibular gland and cervical lymphadenopathy. Suggestive of sialadenitis, Cystic lesion adjacent to the genioglossus muscle complex, deep to the level of the geniohyoid muscle with no superficial extension May represent simple ranula / epidermoid cyst (Fig - 9)

After all clinical and preoperative evaluations excision of ranula along with the sublingual gland was under general anaesthesia. The excised sample was sent for histopathological examination, The given H&E stained soft tissue section shows numerous minor salivary gland acini with connective tissue showing areas of mucin spillage along with chronic inflammatory cell infiltrate predominantly lymphocytes, plasma cells and macrophages are evident suggestive of mucous retention cyst(Fig - 10). Child was under follow up for the past 6 months and there was no recurrence.
DISCUSSION & REVIEW

The name "ranula" is derived from the Latin word “rana” which means "frog" which resembles translucent underbelly/air sac of frog. (Urso-Baiarda et al. 2003; Haberal et al. 2004) Ranula can present clinically in three distinct forms “Sublingual ranula" are most common and presents with intraoral sublingual swelling. The ranula that are located cervically beyond mylohyoid are termed "plunging ranula", and those having an oral and cervical component are called "sublingual plunging ranula". Studies signify that oral ranula tend to occur in children and young adults with peak frequency in the 2nd decade of life. Plunging ranula tend to occur in the older age group and are more infrequent.
Epidemiological data on the same is limited, but the overall prevalence of ranula in the paediatric population is 0.2 cases per 1000. Congenital ranula are rarer, with an incidence of 0.74%, with prenatal diagnosis rarely reported\cite{3}.

Zhao et al., in a review of 580 cases and reported that ranulas are most prevalent in the second decade of life and are slightly more common in females (M/F = 1/1.2) but a distinct male predilection was noted for the plunging ranula (M/F = 1/0.74)\cite{4}. Both the cases reported here are female children.

The etiology is unknown but it has been described in association with congenital anomalies, trauma, and disease of the sublingual gland (Davison et al. 1998). Clinically most ranula are asymptomatic and self-limiting, presenting as a painless, fluctuant, translucent, bluish dome shaped swelling commonly seen in the floor of the mouth\cite{5}.

Hypertension created within the duct due to obstruction leads to acinar rupture in the salivary gland and then extravasation of the mucus. Initially there is traumatic rupture of the excretory duct followed by extravasation and subsequent accumulation of saliva within the tissue, as shown by experimental studies\cite{4,5,6}.

In the submandibular gland, mucus extravasation initiates an inflammatory reaction leading to fibrosis that often prevents further expansion of the pseudocyst. In sublingual gland however, the secretory activity of the gland is persistent and continues despite fibrosis leading to more substantial pseudocyst formation\cite{7}.

A number of differential lesions may be encountered in the floor of the mouth or submandibular space region. They include congenital abnormalities (cystic hygromas, branchial cleft cysts, and thyroglossal duct cysts), benign lesions (epidermoid cysts, dermoid tumors, and lipomas), malignant neoplasia, and other lesions (abscess, mucocele and sarcoidosis). While the most of the mentioned lesions required detailed investigations the diagnosis of ranula is largely clinical (Morita et al. 2003; Urso-Baiarda et al. 2003)\cite{8,9}. In our case, the diagnosis of ranula was investigated using an ultrasound and later confirmed histopathologically.

Ultrasonography, computed tomography, MRI and fine needle aspiration have been shown to be useful when narrowing the potential differentials, however MRI scan is considered a goldstandard investigation; besides giving high resolution images, it determines the precise
location and content of the lesion and enhances\textsuperscript{[7,10]} On an average of 8 pediatric ranula cases reported in our metanalysis, 4 cases the investigation of choice was MRI.

Computed tomography creates a hallmark with its “tail sign” which is pathognomonic for the plunging ranula. This tail is due to extension behind the mylohyoid muscle that confirms the ranula to arise from the sublingual gland and is especially useful in differential diagnosis of cervical ranula\textsuperscript{[8]}.

Biochemical analysis of aspiration fluid reveals high protein and amylase contents. Interestingly congenital lesions can now be diagnosed prenatally by ultrasonography and an EXIT (exuterointrapartum treatment) procedure may be followed for the treatment in such cases\textsuperscript{[11,12]} Manish et al, in 2016 reported a case on congenital ranula treated with EXIT.

Management of ranulas is a polarising topic Multiple options exist, including surveillance, needle aspiration, surgical excision of the cyst, sublingual gland excision along with the cyst, marsupialisation, sclerotherapy, laser excision or cryosurgery\textsuperscript{[13]}. In a review of 580 ranulas, recurrence rates for marsupialization, excision of ranula, and excision of the sublingual gland combined with the lesion were 66.7\%, 57.7\%, and 1.2\%, respectively\textsuperscript{[14]}. Excision of the ranula along with the gland was the treatment of choice in both our cases and it reported with no recurrence after a follow up of 6 months.

The treatment protocol for paediatric ranula is still controversial. Most likely the explanation for resolution would be a rupture as a result of feeding. Four to six Neonatal imperforate ducts may resolve spontaneously with feeding. Hence they should be observed for potential resolution for several months in uncomplicated cases\textsuperscript{[15]} Soni et al, in 2012 reported a case of congenital ranula which resolved spontaneously following therapeutic aspiration of the contents.
<table>
<thead>
<tr>
<th>S.No</th>
<th>Article</th>
<th>Age/Sex</th>
<th>Etiology</th>
<th>Site</th>
<th>Clinical symptoms</th>
<th>Clinical Features</th>
<th>Investigations</th>
<th>Treatment</th>
<th>Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>Gautam et al, 2013, New Delhi</td>
<td>20 days</td>
<td>Congenital</td>
<td>Midline floor of mouth</td>
<td>Difficulty in feeding and Noisy breathing</td>
<td>1 cm size</td>
<td>CT &amp; MRI</td>
<td>Excision of Ranula</td>
<td>No</td>
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<td>2.</td>
<td>Soni et al, 2012,</td>
<td>1 day/F</td>
<td>Congenital</td>
<td>Left side floor of mouth</td>
<td>Asymptomatic</td>
<td>1.5*1cm in size, lifting tongue</td>
<td>None</td>
<td>Aspiration</td>
<td>No</td>
</tr>
<tr>
<td>3.</td>
<td>Sirin et al, 2016, Lebanon</td>
<td>4 mon/F</td>
<td>Congenital</td>
<td>Floor of the mouth</td>
<td>Difficulty in feeding &amp; increase in size of the swelling of,</td>
<td>4× 3cm, elevation of tongue</td>
<td>CT scan</td>
<td>intraoral excision with preservation of Wharton’s ducts</td>
<td>Yes</td>
</tr>
<tr>
<td>4.</td>
<td>Pavai et al, 2010,</td>
<td>8 mon/ F</td>
<td>Unknown</td>
<td>Left side floor of the mouth</td>
<td>-</td>
<td>Involving sublingual region and extending into submandibular, submental region.</td>
<td>MRI</td>
<td>Marsuplization and then Intraoral excision of sublingual gland</td>
<td>Yes</td>
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<td>5.</td>
<td>Pavai et al, 2010,</td>
<td>10 yr/ F</td>
<td>Unknown</td>
<td>Floor of the mouth</td>
<td>-</td>
<td>3 × 4 cm in size</td>
<td>MRI</td>
<td>Marsuplization followed by excision of sublingual gland</td>
<td>Yes</td>
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<td>6.</td>
<td>Sathanantham et al, 2015</td>
<td>7yr/ F</td>
<td>Unknown</td>
<td>Floor of the mouth</td>
<td>Enlarging in nature with airway</td>
<td></td>
<td>MRI</td>
<td>Nasotracheal intubation, following</td>
<td>No</td>
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<td></td>
<td>Authors, Year</td>
<td>Age/Gender</td>
<td>Gender</td>
<td>Location</td>
<td>Description</td>
<td>Imaging</td>
<td>Excision Details</td>
<td>Outcome</td>
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<td>7.</td>
<td>Effat et al, 2012</td>
<td>14yr/M</td>
<td>Unknown</td>
<td>Floor of the mouth</td>
<td>Rapidly enlarging neck swelling</td>
<td>CT-Scan</td>
<td>Nasotracheal intubation, following excision of the cyst</td>
<td>No</td>
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<tr>
<td>8.</td>
<td>Margalit et al, 2016</td>
<td>11yr/F</td>
<td>Unknown</td>
<td>Floor of the mouth</td>
<td>Difficulty in talking and eating. 4.5 X 3 cm sublingual mass; soft, movable and non-tender. The contents had a bluish hue.</td>
<td>Ultrasound</td>
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CONCLUSION
Ranula, an intraoral lesion with a marked clinical appearance creating much concert; is actually a lesion with minimal symptoms if diagnosed and managed at the right time. The fact is more appropriate for the pediatric group. The cases of pediatric ranula reported and recorded in the current review, presented with a alarming clinical appearance but rather ended on a calm noted with very few reported recurrence. Thus we highlight the fact the correct diagnosis, adequate investigations and effective treatment is the most essential aspect in addressing ranula.

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18. Ili Margalit, Point-of-Care Ultrasound to Diagnose a Simple Ranula, Western Journal of Emergency Medicine, November 2016.