

# Sublingual Foregut Cyst Lined by Respiratory Epithelium- A Rare Cause of Cystic Lesion in the Floor of the Mouth

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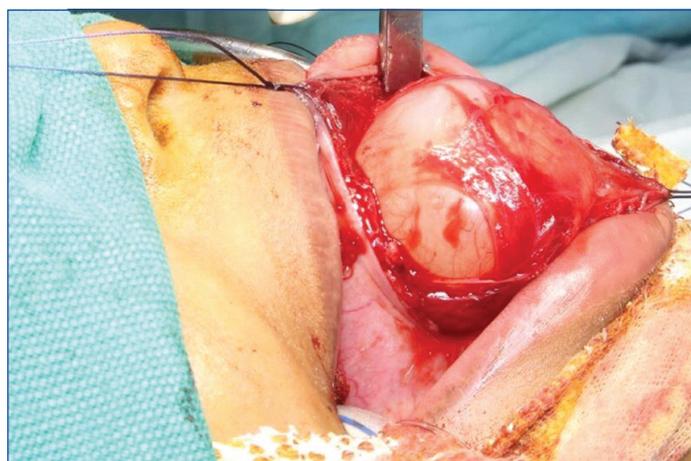
## ABSTRACT

Foregut cysts of the oral cavity are rare developmental anomalies which are lined by gastric, intestinal, squamous or respiratory epithelium or a mixture of these epithelia. A 10-year-old boy presented with a large cyst in the floor of the mouth and had difficulty in speech and chewing of food. On Magnetic Resonance Imaging (MRI) examination a diagnosis of ranula was given. Surgical excision was done and a diagnosis of foregut cyst lined by respiratory epithelium was given on histopathological examination. Till now only 21 cases of lingual foregut cyst with respiratory lining have been reported. We report the 22<sup>nd</sup> case for its rarity and because these lesions can pose a diagnostic dilemma and should be included in the differential diagnosis of cystic lesions in the oral cavity especially in paediatric population.

**Keywords:** Developmental cyst, Pseudostratified ciliated epithelium, Sublingual swelling

## CASE REPORT

A 10-year-old boy presented with a swelling in the floor of the mouth which was present since birth. Initially, the swelling was small and it gradually increased to the present size. He had difficulty in speaking since an early age and chewing of solid food since one month. His medical history was unremarkable. On clinical examination, the swelling was non-tender, soft, cystic, translucent, and measured 5×3.52 cm in size. The tongue was elevated due to the swelling [Table/Fig-1]. On MRI, a well-defined cystic, non-enhancing sublingual lesion was seen in the floor of the mouth reaching up to the mylohyoid muscle, and a diagnosis suggestive of ranula was given. Under general anaesthesia, an incision was taken extending from the tip of the tongue till the posterior tongue in the sublingual region and the cyst was enucleated by blunt dissection and the cyst was separated from the intrinsic muscles of the tongue [Table/Fig-2].



[Table/Fig-2]: Intraoperative dissection and excision of the cyst.



[Table/Fig-1]: Clinical picture showing a cystic swelling in the floor of the mouth.



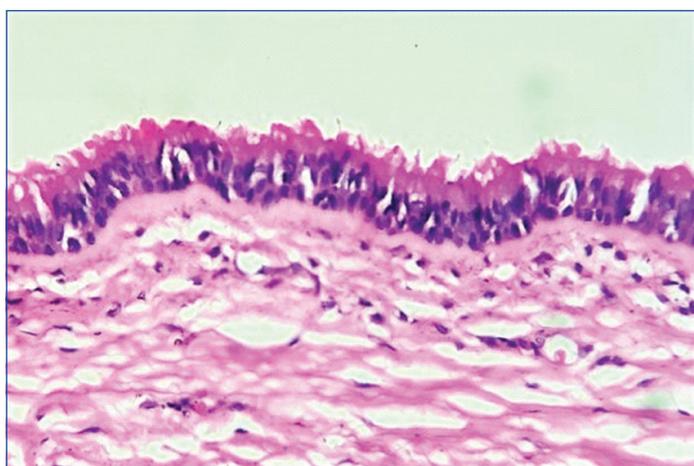
[Table/Fig-3]: Gross specimen showing a large smooth walled cyst.

Grossly, a large single cystic mass was seen measuring 6×4×3 cm [Table/Fig-3]. On sectioning, the cyst had a smooth wall, was uniloculated, and filled with mucoid material. Microscopic examination of the cyst wall showed a cyst lined by pseudostratified ciliated epithelium which was flattened at places [Table/Fig-4]. Wall of the cyst showed fibro collagenous tissue along with underlying muscle. A diagnosis of benign foregut cyst with respiratory lining was given. The postoperative course was uneventful.

## DISCUSSION

Foregut cysts are a rare cause of cystic lesions involving the tongue or the floor of the mouth. They have a varied terminology and were also known as lingual choristoma, enteric duplication cysts, lingual alimentary cysts etc., [1]. The commonest location of these cysts is in the chest and abdomen but involvement of tongue and oral cavity is unusual and is seen only in 0.3% of the cases [1,2]. There are three criteria to make a diagnosis of foregut

cysts- have to contain epithelium derived from foregut, should be attached to a portion of the foregut and covered by smooth muscle layer [2]. The epithelial lining seen in these cysts is gastric, intestinal, respiratory or a mixture of these cells which suggests a foregut origin [1]. Foregut cysts of oral cavity are usually present at birth and gradually increase in size. A few cases have been detected during antenatal ultrasound examination [3]. Most cases present as asymptomatic swelling in the floor of the mouth but in 30% of cases they present with difficulty in feeding, swallowing, respiration or speech [4]. The present case presented with difficulty in speech and chewing of solid food because of its large size and location in the floor of the mouth. Peptic ulceration in cyst with gastric lining, sinus formation with chronic mucus secretion have been reported. Malignant transformation has been reported in a long standing case of foregut cyst of head and neck [2]. MRI is recommended to delineate the extent of these cysts but it shows overlapping features with more common cystic lesions occurring at this location [5].



**[Table/Fig-4]:** Microscopy of the cyst showing respiratory lining composed of ciliated pseudo- stratified epithelium (H&E 40X).

Manor Y et al., have histologically reviewed 52 cases of lingual cysts out of which only three involved the floor of the mouth [6]. They reported one case of lingual cyst on the tongue lined by respiratory epithelium. Age ranged from new born to 42 years with a mean age of 5 1/2 years. Cysts were more common in males as compared to females with a male female ratio of 1.6:1. Grossly the size varied from 1 cm to 6.5 cm with a mean size of 2.3 cm. Out of the 52 cases of foregut cyst reviewed histologically, 12 contained mainly respiratory epithelium, 21 cases contained mainly gastric epithelium, three cases contained mainly intestinal epithelium, one contained both gastric and intestinal epithelium and 15 were composed of combination of respiratory and gastric or intestinal epithelium. As these cysts were reported by different names Manor recommended a uniform nomenclature with a descriptive diagnosis based on histopathological examination. He referred them as foregut cysts with gastric epithelium, foregut cysts with respiratory epithelium and foregut cysts with respiratory and gastric epithelium [6]. Peters SM et al., have reported 2 cases of foregut cysts lined with respiratory epithelium which is similar to our case [7]. On review of literature they have found only 19 similar cases reported earlier which had only respiratory lining with absence of gastric epithelium. Patients presented with swelling on the dorsal tongue, ventral tongue and floor of the mouth [8-17]. The present case is thus the 22<sup>nd</sup> case to be reported [Table/Fig-5].

There are many overlapping theories for the development of these lesions; hence the exact pathogenesis remains debatable. They could arise from developmental derangement of the foregut. During the 3<sup>rd</sup> week of embryogenesis, the ventral part of the foregut gives rise to respiratory tract and the dorsal part to proximal gastrointestinal tract. The pharyngeal arches lie at proximity to the primitive foregut; hence the pluripotent cells of the foregut may sometimes get entrapped between the developing tongue and when exposed to varying influences and environment differentiate into respiratory or gastro-intestinal epithelium [2, 18].

Cystic lesions in the floor of the mouth have a wide range of differential diagnosis and include ranula, dermoid cyst, thyroglossal

| Sr. No. | Author (Year)                     | No. of cases | Age (Years)               | Sex    | Site                                      | Clinical symptoms                         | Histopathologic features (Cyst lining)  |
|---------|-----------------------------------|--------------|---------------------------|--------|---|---|---|
| 1.      | Fink (1963) [8]                   | 1            | 5                         | Male   | Dorsum tongue, anterior third             | Difficulty in eating and drinking.        | Pseudostratified ciliated columnar+cuboidal epithelium                                |
| 2.      | Constantinides et al., (1982) [9] | 1            | 9 month old               | Female | Ventral tongue                            | Difficulty in eating and closing of mouth | Pseudostratified ciliated columnar+squamous+cuboidal epithelium                       |
| 3.      | Wiersma et al., (1992) [10]       | 7            | Various (neonate-6 years) |        | Anterior two-thirds of tongue             | Asymptomatic; Difficulty in feeding       | Pseudostratified ciliated columnar+squamous +cuboidal+columnar epithelium             |
| 4.      | Naidoo (1997) [11]                | 1            | 42                        | Male   | Centre of dorsum of tongue                | Difficulty in eating and speaking         | Pseudostratified non-ciliated columnar+squamous+cuboidal+ciliated columnar epithelium |
| 5.      | Kim et al., (1998) [12]           | 1            | 27                        | Male   | Sublingual                                | Non-specific                              | Pseudostratified ciliated columnar+squamous epithelium                                |
| 6.      | Manor Y et al., [6]               | 1            | 11                        | Male   | Body of tongue                            | Difficulty in speaking and swallowing     | Pseudostratified ciliated columnar+cuboidal epithelium+goblet cells                   |
| 7.      | Azanero (2009) [13]               | 2            | 4                         | Male   | Right ventral tongue                      | Difficulty in breastfeeding               | Pseudostratified ciliated columnar+squamous epithelium+goblet cells                   |
|         |                                   |              | 21                        | Male   | Anterior middle third of dorsum of tongue | Difficulty in eating and speaking         | Pseudostratified ciliated columnar+squamous epithelium+goblet cells                   |
| 8.      | Boffano P et al., [14]            | 1            | 35                        | Female | Floor of mouth                            | Asymptomatic                              | Pseudostratified ciliated columnar+goblet cells                                       |
| 9.      | Chai RL et al., [5]               | 1            | 6 month old               | Female | Ventral tongue                            | Difficulty in feeding                     | Pseudostratified ciliated columnar  |
| 10.     | Junejo et al., (2011) [15]        | 1            | 1                         | Female | Anterior dorsal tongue                    | Difficulty in eating                      | Pseudostratified ciliated columnar+squamous epithelium                                |
| 11.     | Fortier et al., (2013) [16]       | 1            | 17 month old              | Male   | Left anterior tongue                      | Asymptomatic                              | Pseudostratified ciliated columnar+some mucinous glands                               |
| 12.     | Kwak EJ et al., [17]              | 1            | 2                         | Female | Ventral tongue                            | Asymptomatic                              | Pseudostratified ciliated columnar  |
| 13.     | Peters SM et al., [7]             | 2            | 10                        | Male   | Floor of mouth                            | Asymptomatic                              | Ciliated columnar   |
|         |                                   |              | 27                        | Female | Floor of mouth                            | Asymptomatic                              | Pseudostratified ciliated columnar+goblet cells                                       |
| 14.     | Current case                      | 1            | 10                        | Male   | Floor of mouth                            | Difficulty eating and speaking            | Pseudostratified ciliated columnar  |

**[Table/Fig-5]:** Cases of lingual cyst lined by respiratory epithelium with absence of gastric epithelium.

duct cyst, lymphangioma, haemangioma, lymphoepithelial cyst and salivary gland neoplasms [5,14,17]. Clinically it is difficult to differentiate among these lesions. Diagnosis is made after excision and histopathological examination. Oral cysts lined by respiratory epithelium are benign lesions and are treated by surgical excision.

## CONCLUSION

Lingual cysts lined by respiratory epithelium are a rare cause of cystic lesion in the floor of the mouth with only 21 cases reported in literature. Early diagnosis and excision of foregut cyst is necessary because it can cause difficulty in feeding, speech, swallowing and respiration. As foregut cysts have a varied terminology it is recommended to label it as a foregut cyst followed by the type of epithelial lining.

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