

Case Report

A Rare Case Report of Oral Lymphangioma in Buccal Mucosa

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Abstract

Lymphangiomas are rare benign lymphatic abnormalities which mostly occur in the head and neck region and in mouth the preferred area of involvement is the anterior two-thirds of dorsal tongue. Here an acquired oral lymphangioma of the buccal mucosa in a 50-year-old woman complaining of the presence of a mass in her mouth is presented. Pathologic examination of the specimen revealed the diagnosis of lymphangioma. A review of the PUBMED indexed papers in English was carried out and the results were discussed.

Keywords: Lymphangioma; Mouth mucosa; Retromolar pad

Introduction

Lymphangiomas are rare, benign lesions which can be congenital or acquired. In the 50% of cases, lymphangioma can be seen at birth and up to 90% of lymphangiomas are developed before 20 years of age [1,2].

Lymphangiomas arise in the head and neck region in more than 90% of the cases. Occurrence in the oral cavity is rare but in case of occurrence in the mouth, they usually involve dorsum of tongue more often. Involvement of buccal mucosa and retro molar region are rare [2,3].

The main purpose of managing lymphangioma is to restore or maintain function and esthetic of the affected area [4]. Treatment of these lesions consists of various methods, such as close follow ups, laser therapy, cryotherapy, electrocautery, sclerotherapy and surgical excision [5].

The following report is about a rare case of oral lymphangioma in the buccal mucosa and retro molar region.

Case Presentation

A 50-year-old female patient is presented with the chief complaint of a lump sized as a walnut on the left cheek existing for 2 years which has shrunk after extraction of 2 posterior teeth.

Gross examination revealed a 1 cm × 1.5 cm nodule with pebbly surface on the left buccal mucosa extending to the retromolar region.

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The incisional biopsy was done for nodular portion of the lesion under local anesthesia. The specimen examined using Hematoxylin and Eosin (H&E) staining and the lesion diagnosed as lymphangioma (Figure 1).

Discussion

Lymphangiomas are benign, vascular anomalies with lymphatic differentiation. There is doubt whether these lesions are neoplastic, hamartomatous, or malformations. Lymphangiomas are most seen in the head and neck area, followed by limbs, trunk and buttocks. Intraoral lymphangiomas mostly occur in anterior two-thirds of the tongue, lips, floor of mouth, palate and buccal mucosa. Also they may be seen in larynx, parotid gland and tonsils. It is reported that 50% of the lymphangiomas are seen at birth with no previous familial history [1,2,6,7].

A search of Lymphangioma in PUBMED and GOOGLE SCHOLAR found 209 cases of oral lymphangiomas in English literature up to May 2020. The table lists country, age of first notice, age of management, location, diameter, chief complaint, treatment method and recurrence, from the reported 210 cases of lymphangioma (209 previous cases and our case), 104 cases were seen in male and 96 in female (m/f ratio is 1.08) with the mean and median age of 9.42 and 2 respectively, ranging from birth to 78 years old. The lesions varied in size from 0.3 cm to 10 cm in the greatest diameter.

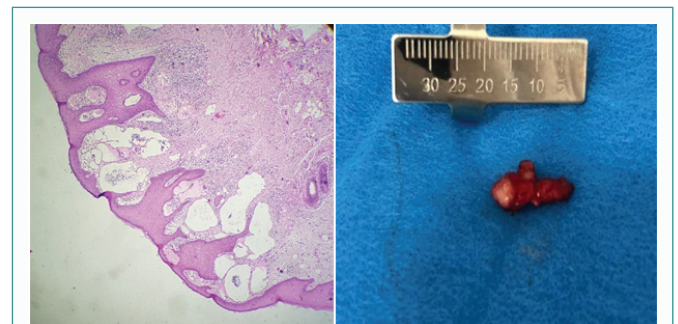


Figure 1: The lesion regressed after biopsy and therefore, no action for treatment was done.

Various theories have been proposed for the pathogenesis of lymphangiomas which are mentioned as follows: The first theory suggests that the reason for arise of congenital lymphangioma, is the sequestration of primary lymphatic channels and subsequent inability to join main lymphatic vessels or veins. The second theory is that the inability of lymphatic channels to drain into the veins leads to lymphectasia and lymph stasis, development of blind, dilated lymphatic sacs [1]. The third theory explains that lymphangiomas can form through abnormal budding of lymphatic vessels and forming of new atypical branches. The fourth theory proposes that chronic inflammation can be the cause of lymphangioma by stimulating the growth of lymphatic vessels. The last and newest theory demonstrates that the expression of 2 growth factors called Vascular Endothelial Growth Factor C (VEGF-C) and vascular endothelial growth factor R3, are increased in lymphangiomas, while the expression of 2 angiogenesis inhibitors named pigment epithelium derived factor and thrombospondin-1 may be reduced [2].

Clinical manifestations of lymphangiomas usually include plaques consist of thin walled, translucent vesicles that look like frog eggs. Occasionally vesicles would be seen in a range of red to purple, suggesting the hemorrhage into the lymphatic channels. Small sized lymphangiomas are usually asymptomatic, while large ones can interfere with functions like speech or mastication. Rarely, they can be dangerous by obstructing airway, secondary infection and subsequent cellulitis [1,7].

From the histopathological aspect, lymphangiomas are made up of fluid-filled lymphatic channels and they are subdivided to 4 groups according to the size:

- Capillary lymphangioma: (lymphangioma simplex, microcystic lymphangioma, lymphangioma circumscriptum) that occurs in cutaneous, subcutaneous or infrequently muscular tissue.
- Cavernous lymphangioma (macrocytic lymphangioma): that can infiltrate into the adjacent tissues.
- Cystic lymphangioma (cystic hygroma): which is characterized by cystic spaces and epithelium lining.
- Lymphangiomasarcoma: an uncommon malignant neoplasm.
- However, the classification cannot set a clear border between different types and subsequently the appropriate treatment plan.

The lymphatic spaces contain lymphatic fluid and lymphocytes. Occasionally, red blood cells can be found due to trauma and following hemorrhage [1,7-9].

Since endothelial cells express CD31 and D 2-40, lymphangiomas are reactive to these two Immunohistochemical markers. CD34 expression in endothelial cells is controversial. Alrashdan et al. [1] declared the negative result of CD34 marker reactivity; however Nelson et al believed that lymphangiomas' endothelial cells express CD34 [1,7].

Differential diagnosis of oral lymphangioma includes granular cell tumor, hemangioma, pyogenic granuloma, squamous papilloma, epidermal inclusion cyst, juvenile angiofibroma, lipoma, adenoma, chondroma, vascular malformations and lingual thyroid [2].

The aim of treatment is to solve complications such as pain, esthetic problems, functional impairment and enlargement. Therefore, due to the benign nature of lymphangioma, if any of the problems mentioned above do not exist, it can just be followed [4]. Spontaneous remission of the lesion has been reported as well [10]. This is more likely to occur in infants than adults [11].

Several treatment methods have been proposed such as: surgical excision, aspiration or drainage, radiotherapy, radiofrequency ablation, sclerotherapy and cryotherapy [12].

Surgical excision is the classic and common treatment method, whereas infiltration into the adjacent tissues increases the risk of recurrence through lack of precision to excise the whole lesion. Complications after surgery may be hemorrhage, fibrosis, lymph discharge, vital structures damage, nerve damage and wound infection [6,12].

Aspiration and drainage are temporary treatments for reducing the size or decompression rather than permanent treatments [12].

Radiotherapy was used in the past for management of lymphangioma but nowadays it is not an acceptable method because of inability to completely eliminate the lesion, post radiation malignancy and damage to the adjacent tissues. We found 2 cases of post radiation malignant neoplasms (angiosarcoma, squamous cell carcinoma) following radiotherapy of lymphangioma in the literature [13,14].

Sclerotherapy is another treatment method that has acceptable results. Bleomycin, OK432, doxycycline, tetracycline, steroids, cyclophosphamide and hypertonic saline are used as sclerosing agents. Post procedure complications of using bleomycin are pulmonary toxicity, erythema, edema and fever. To prevent these complications combination of steroids and bleomycin is recommended. Although sclerotherapy doesn't seem to have effect on macrocystic or mixed lymphangiomas, doxycycline seems to be effective in these cases [4,6,9].

Radiofrequency ablation doesn't have the complications like injury, infection, bleeding, inflammation and edema. This can be done in two ways: low frequency or high frequency devices. Both are used for localized superficial lesions but the higher the frequency, the deeper the tissue destruction. Deeper tissue destruction also results in tissue fibrosis [12,15].

Cryotherapy is the removal of a lesion using extreme cold. This is done by the process of frosting/defrosting the lesion in 20 seconds to 2 minute cycles using a cryoprobe. The advantages of cryotherapy are maintaining esthetics, reduced formation of fibrous tissue, diminished bleeding and preserving surrounding vital structures; while the complications are swelling and denaturing adjacent tissues which occur as a result of uncontrolled use (Table 1) [8].

Recurrence is common in lymphangiomas because of incomplete encapsulation and infiltrative nature. A recurrence rate of 15% to 35% has been reported in articles (in our findings it was 24.24%, 24 of 99 cases) [2,7].

References

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Table 1: Cases Recurrence.

Case	Author	Year	Country	Gender	Age (procedure)	Age (first seen)	Site	Size (cm)	Chief complaint	Treatment	Recurrence
1	Pati [16]	-	India	Male	11	11	Right buccal mucosa	3 × 2.5 × 1	Swelling	Follow up	-
2	Carling [17]	1910	UK	Female	13	13	Tongue	-	-	-	-
3	Weber [18]	1910	-	Female	22	birth	Left parotid gland	2	-	-	-
4	Odgers [19]	1913	UK	Male	29	birth	Tongue	-	Pain-Bleeding	Excision	-
5	Howell [20]	1919	UK	Male	6	3	Tongue	-	Ulcers-Pain	Radium therapy	-
6	Whitfield [21]	1921	UK	Female	10	11	Tongue	-	-	-	-
7	Petit [22]	1923	UK	Male	-	-	Tongue	-	pain	-	-
8	Woolf [23]	1927	UK	Male	5 months	birth	Tongue	-	-	-	-
9	Wakeley [24]	1928	UK	-	8	3	Tongue	-	Swelling	Lingual artery ligation- Radium therapy- X-ray therapy	-
10	Klaber [25]	1936	UK	Male	6	6	Tongue	-	-	Radiotherapy	-
11	Christensen [26]	1953	USA	Male	20	birth	Tongue	3.8 × 2.5 × 1.3	Difficulty with speech- Dysphagia	Excision	no
12	Robinson [27]	1953	Uk	Female	1	birth	Tongue	7 × 1.5	Enlargement- Protrusion	Radiotherapy- Partial glossectomy	yes
13				Male	6	birth	Tongue	-	Enlargement- Ulcers	Radiotherapy	no
14				Male	8 Months	birth	Tongue	-	Enlargement	Radiotherapy	no
15				Male	3	2	Tongue	-	Enlargement	X-ray therapy	no
16				Female	10	birth	Tongue	2	Enlargement	X-ray therapy- Excision	no
17	Robinson [28]	1959	USA	Female	5 months	birth	Tongue, sub mental region, submandibular region, Left cheek	10 × 6 × 3	Difficulty with occlusion- Asymmetry	Partial resection	-
18	Harrison [29]	1960	USA	Female	27	27	Right tonsil	1.4 × 0.8 × 0.8-1 × 0.7 × 0.7	Sore throat- Bleeding	Tonsillectomy	-
19	Morfit [30]	1960	USA	Male	4	Birth	Tongue- Sub mental, Sub maxillary, Left parotid region	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy- Parotidectomy	-
20				Female	8	Birth	Tongue- Soft palate- floor of mouth- bilateral sub mental, parotid region	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy- Parotidectomy	-
21				Female	8	Birth	Tongue	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy	-
22				Female	3	3	Tongue	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy	-
23				Male	4	1	Tongue- Floor of mouth	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy	-
24				Male	6	2	Tongue- Floor of mouth	-	Enlargement- Ulcers	Partial glossectomy	-
25				Male	4	1	Tongue	-	Enlargement- Ulcers	Partial glossectomy-	-
26				Female	2	birth	Tongue- Floor of mouth	-	Enlargement- Ulcers	Partial glossectomy- Sclerotherapy	yes
27	Koop [31]	1961	USA	Male	2 days	Birth	Left Parotid region- Tongue	-	Swelling	Excision- Electrodesiccation	yes
28				Male	3	3	Tongue- Floor of mouth	1.5	Enlargement- Mass existence	Excision- Electrodesiccation	yes
29				Female	6	birth	Tongue	-	Swelling	Excision	-
30				Male	2 weeks	birth	Tongue- Submandibular region	-	Swelling- Difficulty with breathing	Excision- Radiotherapy	yes
31				Female	2 weeks	birth	Tongue- Neck- Sub mental region- Larynx	-	Swelling- Dysphagia	Excision- Electrodesiccation	yes

32				Female	7 months	birth	Tongue- Neck- Floor of mouth	-	Swelling- Difficulty with breathing	Excision	yes
33				Female	2	2	Tongue- Neck	-	Enlargement	Excision- Electrodesiccation- Radiotherapy	yes
34				Male	6 Weeks	birth	Tongue- Submandibular area	-	Mass existence- Enlargement	Excision- Radiotherapy	no
35	Goldman [32]	1963	USA	Female	11 months	birth	Left parotid gland	5 × 3 × 1.5-4 × 3 × 2	Mass existence	Excision	no
36	Kabelka [33]	1966	-	Male	3	birth	Tongue	4	Enlargement	Resection	no
37	Visvanathan [34]	1971	Australia	Male	31	31	Left Tonsil	2.5 × 1.3 × 1.2	Dysphagia- Sore throat- Hearing impairment	Tonsillectomy	-
38	Kornblut [35]	1973	Germany	Male	3	birth	Left Parotid	-	Swelling	Parotidectomy	yes
39				Male	3.5	birth	Right parotid	-	Swelling	Parotidectomy	-
40	Brandrup [36]	1976	Denmark	Male	13	1	Tongue	-	Difficulty with mouth closing	Electrodesiccation- Radium therapy- Excision	-
41	Mendex [37]	1976	-	Female	18	-	Tongue-Neck	-	Enlargement	Resection	-
42	Marshall [38]	1977	USA	Male	8	birth	Tongue, Soft palate, Floor of mouth	-	Impaired occlusion, tongue enlargement	resection	-
43	Nagata [13]	1984	Japan	Male	27	5	Right Buccal mucosa- Lower lip	-	Mass existence	Excision	yes
44	Takato [39]	1984	Japan	Female	5	birth	Right parotid	4.3	Swelling	Subtotal parotidectomy	-
45	Al Samarrae [40]	1985	Jordan	Male	35	29	Left tonsil	2.5 × 1.5	Mass existence- Sore throat	Tonsillectomy	-
46				Female	35	-	Left tonsil	2.8 × 1.5	Headache- Tinnitus- Dysphagia	Tonsillectomy	-
47	Eppley [41]	1985	USA	Female	19	17	upper lip	0.6	Swelling	Excision	no
48	Rice [42]	1985	USA	Male	5	birth	Tongue	-	Swelling- pain	Resection	yes
49	Ikemura [43]	1987	Japan	Male	1	birth	Tongue extended to neck	-	Swelling	Resection- Radio therapy	no
50	Balakrishnan [44]	1991	London	Female	8	birth	Tongue- Submandibular region- Floor of mouth	-	Swelling- Airway obstruction	Surgical Excision- Laser therapy	yes
51				Male	8	2 weeks	Tongue- Neck- Floor of mouth- Soft palate- Tonsils- Epiglottis- Supraglottis	-	Difficulty with breathing- Swelling	Surgical Excision- Laser therapy	yes
52				Male	5	birth	Neck- Bilateral parotid regions- Floor of mouth- Tongue	-	Swelling	Laser therapy- Resection	yes
53				Female	5	birth	Tongue- Neck- Floor of mouth- Epiglottis	-	Swelling	Laser therapy- Resection	yes
54				Female	3	6 months	Tongue- Floor of mouth	-	Swelling- Bleeding	Laser therapy- Resection	no
55				Male	8	birth	Neck- Left buccal mucosa- tongue- Floor of mouth- Palate	-	Dysphagia- Nerve palsy- Swelling	Resection- Laser therapy	yes
56				male	4	1 week	Neck- Submandibular region- Bilateral buccal mucosa- right parotid region- both lips- Tongue	-	Swelling- difficulty with breathing	Laser therapy	yes

57	Basu [45]	1992	India	Male	3 days	birth	Tongue	7.5*2	Tongue enlargement	Aspiration	yes
58				Female	5	2	Tongue		Difficulty with mouth closing-Difficulty with speech	Partial glossectomy	no
59				Male	5 months	birth	Tongue	2 × 2	Swelling	Excision	no
60	Livesey [46]	1992	UK	Male	26	3	Parotid gland	10 × 6	Swelling	Excision	no
61	Lobitz [47]	1995	USA	Female	6	6	Tongue	2 × 4	Mass Existence	Excision	no
62	Hazra [48]	1996	India	Female	38	38	Tonsil	4.5 × 2	Mass feeling-Sore throat	Tonsillectomy	no
63	Tsui [49]	1996	China	Male	10	10	Left parotid gland	5 × 5.5	Swelling, pain	Antibiotic therapy	-
64	Morgan [50]	1997	UK	Male	35	35	Left parotid gland-Submandibular gland	4 × 4	Mass existence	Total parotidectomy-resection	no
65	Paladini [51]	1998	Italy	-	Prenatal(33 weeks)	prenatal	Tongue- Floor of mouth	3.5 × 3	-	Immediate Cesarean by 38 th week- Drainage- Excision	no
66	Rao [52]	1998	India	Female	25	birth	Tongue- Right temporal region-submandibular region	5 × 5	Swelling	-	-
67	Tasca [53]	1999	Australia	Female	6	6	Tongue	-	Sore throat, Swelling	Antibiotic therapy	-
68	daLilly-Tariah [54]	2000	Nigeria	Female	22	21	Right parotid gland	5.5 × 4.6 × 2.5	Swelling	Subtotal parotidectomy	-
69	Henke [55]	2001	USA	Male	34	34	Left parotid gland	3 × 5	Swelling	Excision	no
70	Berry [14]	2002	USA	Female	64	19	Tongue	2.8 × 3	Dysphagia-Sore throat-Earache	Drainage- Excision- Radiotherapy- Chemotherapy	-
71	Park [56]	2002	Korea	Female	69	39	Mandible- Lower lip	4 × 2 × 1	Irritation-Discoloration	-	-
72	Guelmann [57]	2003	USA	Female	4	birth	Tongue	-	Enlargement	Sclerotherapy	-
73	Lamaroon [58]	2003	Thailand	Female	11	birth	Tongue	-	Pain	Follow up	-
74	Tei [59]	2003	Japan	Female	5	birth	Tongue	6 × 5	Enlargement-Difficulty with mouth closing	Partial Glossectomy	no
75	Boardman [60]	2004	Australia	Male	22	22	Left tonsil	-	-	Bilateral tonsillectomy	-
76	Mandel [11]	2004	USA	Male	21	4	Left parotid gland	-	Swelling	Excision	-
77	Poh [61]	2005	Canada	Female	61	61	Tongue- Floor of mouth, Left Alveolar ridge, Left Buccal mucosa	6 × 6.5	-	Follow up	-
78				Male	43	43	Tongue	1.8 × 1.2	-	Follow up	-
79				Male	59	59	Mandibular alveolar ridge	1.3 × 0.6	-	Follow up	-
80				Male	42	42	Mandibular alveolar ridge	2.8 × 0.5	-	Follow up	-
81				Female	57	57	Mandibular alveolar ridge	1.5 × 0.5	-	Follow up	-
82				Male	71	71	Maxillary alveolar ridge		-	Follow up	-
83				Male	59	59	Mandibular alveolar ridge		-	Follow up	-
84				Male	78	78	Maxillary alveolar ridge		-	Follow up	-
85				Male	57	57	Maxillary alveolar ridge		-	Follow up	-
86	Raha [62]	2005	India	Male	8	5	Left Tonsil	5 × 3 × 1	Swelling-Difficulty with breathing	Bilateral tonsillectomy	-
87	Rathan [63]	2005	India	Male	5	-	Tongue- Neck	4.7 × 3 × 4	Pain in tooth	-	-
88	Bansal [64]	2006	India	Female	7	birth	Tongue	-	Bleeding-Pain-Dysphagia	-	-

89	Bozkaya [65]	2006	Turkey	Male	21	birth	Buccal mucosa	$3 \times 2.5 \times 1.5$	Pain in tooth-Halitosis	Radiofrequency ablation	no
90	de Queiroz [66]	2006	Brazil	Female	5	2	Tongue	-	Enlargement-Protrusion	Interferon alpha 2a therapy	-
91	Ikedo [67]	2006	Japan	Male	75	48	Tongue	$9 \times 8.5 \times 3$	Difficulty with speaking	Excision	no
92	Kalpidis [68]	2006	Greece	Female	16		Gingiva	0.9	Swelling	Excision	no
93	Khurana [69]	2006	India	Male	10	5	Tongue	-	Bleeding	NSAID Injection	no
94	Purohit [70]	2006	India	Female	8	3months	Tongue	-	Difficulty with speech-dysphagia-Difficulty with breathing	Excision	-
95	Stanescu [71]	2006	Romania	-	9	1	Tongue	-	Pain	-	
96	Leboulanger [72]	2008	France	Female	1	birth	Tongue- Vallecula-Epiglottis	-	Bleeding	Radiofrequency ablation	yes
97				Male	4 months	birth	Tongue- Floor of mouth- Vallecula	-	Swelling	Medication therapy-Radiofrequency ablation-Sclerotherapy	no
98				Female	10	birth	Tongue	-	Bleeding-Difficulty with speaking	Radiofrequency ablation	yes
99				Female	8	birth	Tongue- Submandibular region	-	Truma-Bleeding	Radiofrequency ablation	no
100				Female	6	-	Tongue	-	Mass Existence	Radiofrequency ablation	no
101				Male	14	birth	Tongue	-	Bleeding	Radiofrequency ablation	no
102				Female	7	-	Tongue	-	Enlargement	Radiofrequency ablation	yes
103	Nagaoka [73]	2008	Japan	Female	9	2	Lower lip	$2.8 \times 1.7 \times 0.1$	Swelling	Excision	-
104	FitzGerald [10]	2009	Ireland	Male	5 Months	2 months	Mandibular alveolar ridge	$0.6-0.3 \times 0.4$	Enlargement	Follow up	no
105				Male	4 Weeks	-	Maxillary alveolar ridge crest –mandibular alveolar ridge	0.6	Swelling	Follow up	no
106	Hong [74]	2009	Korea	Male	4	birth	Tongue	-	Enlargement	Sclerotherapy	no
107	Kataria	2009	India	Male	19	8	Tongue	3	Mass Existence-Bleeding-Enlargement	Sclerotherapy	no
108	Tewari [75]	2009	India	Female	13	12	Tongue	-	Enlargement-Dysphagia	Partial Glossectomy-Sclerotherapy	-
109	Dogan [76]	2010	Turkey	Male	35	34	Left buccal mucosa	$3 \times 2 \times 2$	Swelling	Surgical ablation	yes
110	Kotrashetti [77]	2010	India	Female	65	65	Hard palate	1×1.5	Mass existence	Excision	no
111	Ferrara [78]	2010	Italy	Female	7	-	Tongue	-	Abdominal pain	Laser therapy	no
112	Balatsouras [79]	2011	Greece	Male	17	-	Right tonsil	$2.6 \times 1 \times 0.8$	Dysphagia	Tonsillectomy	-
113	Bonet-Coloma [80]	2011	Spain	Female	4	2	Tongue	1.5×1	-	Excision	No
114				Female	14	3	Tongue	1×1	-	Excision	yes
115				Female	8	1	Tongue	0.5×0.5	-	Excision	No
116				Male	11	2	Tongue	1×1	-	Excision	No
117				Male	17 Months	7 months	Tongue	1×1	-	Excision	No
118				Male	13	1	Tongue	1.5×1	-	Excision	Yes
119				Female	3 Months	1 month	Parotid gland	3.6×3	-	Excision	No
120				Female	1 month	birth	Parotid gland	3×2.5	-	Excision	No
121				Female	3	2 months	Floor of mouth	1.5×1.5	-	Excision	No
122				Male	3	1	Tongue	4×4	-	Observation	No
123				Male	2	1month	Tongue	2×2	-	Observation	No

124				Male	2	1	Parotid gland	2 × 2	-	Observation	No
125				Female	15 Months	birth	Buccal mucosa	5 × 3	-	Observation	No
126	Goswami [81]	2011	India	Female	5	birth	Tongue	2 × 2 × 1.5	Enlargement	Surgical Excision	no
127	Kheur [82]	2011	India	Male	13	8	Tongue	-	Pain- Sense of burning	-	-
128	Leboulanger [83]	2011	France	Female	3	1	Tongue-Submandibular region	-	Bleeding	Radiofrequency ablation- Steroid injection- Propranolol therapy	no
129				-	2	-	Tongue	-	Bleeding	Propranolol therapy	No
130				-	11	-	Tongue	-	Bleeding	Propranolol therapy	-
131				-	9	-	Tongue	-	Esthetic problems	Propranolol therapy	-
132	Puricelli [84]	2011	Brazil	Male	3	1	Tongue	1.5 × 1.5	Mass Existence-Pain	Excision	no
133	Vasconcelos [85]	2011	Brazil	Female	17	16	Soft palate	-	Mass Existence-Bleeding	Enucleation	no
134	Alghonaim [15]	2012	Canada	Male	6	6	Tongue	4	Enlargement	Surgical excision- Radiofrequency ablation	no
135	Bansal [86]	2012	India	Female	3	birth	Tongue	1.5 × 2	Swelling- Dysphagia- Difficulty with breathing	Excision	-
136	Bhalla [87]	2012	India	Male	6	-	Tongue	-	Enlargement- Dysphagia- Drooling- Difficulty with breathing	Surgical Excision	-
137	Chung [88]	2012	Korea	Female	3	-	Tongue	-	Enlargement- Dysphagia- Drooling- Difficulty with breathing	Surgical excision	no
138	Jha [89]	2012	India	Male	13	5	Lower lip	-	Enlargement	Excision	no
139	Satar [90]	2012	Turkey	Male	21	-	Right Parotid region and Ramus	2 × 2	Mass existence- Earache- Hearing loss	-	-
140	Sunil [91]	2012	India	Female	7	birth	Tongue	-	Dysphagia- Bleeding	-	-
141				Female	32	32	Tongue	3 × 2	Swelling- Bleeding	-	-
142				Male	8	-	Tongue	-	Bleeding- Enlargement	-	-
143	Bindhu [92]	2013	India	Male	13	13	Lower lip	1.5 × 1	Enlargement	Surgical excision	-
144	Chakravarthy [93]	2013	India	Male	9	9	Tongue	1 × 1	Enlargement	Excision	-
145	Chakravarti [9]	2013	India	Female	5	2 months	Tongue	2 × 2	Swelling- Bleeding after trauma	Sclerotherapy	no
146				Male	4	1 month	Tongue	-	Swelling- Bleeding after trauma	Sclerotherapy	no
147	Ganesh [94]	2013	India	Female	29	12	Tongue	2.3 × 3.3 × 2.4	Enlargement	-	-
148	Haranal [95]	2013	India	Female	28	-	Right buccal mucosa	2 × 1	Swelling	Surgical Excision	-
149	Mardekian [2]	2013	USA	Male	17	-	Left tonsil	3.2	Mass Existence	-	-
150	Torezan [12]	2013	Brasil	Female	18	3	Tongue	-	Bleeding after trauma	Laser Therapy	-
151	Martins [96]	2014	-	Male	6	-	Tongue	-	Mass Existence	-	-
152	Medeiros [97]	2014	-	Female	11	3	Tongue	2.5 × 1.5	Mass existence- Difficulty with speech	Surgical excision	-

153	Akyuz [98]	2014	Turkey	Male	10	birth	Tongue	5 × 6	Swelling- Pain- Dysphagia- Halitosis	Sirolimus therapy	no
154	Berri [99]	2014	Algeria	Female	66	8	Right parotid gland	15 × 13	Enlargement	Surgical Enucleation	-
155	Devi [100]	2014	India	Female	-	-	Soft palate	3 × 2	Swelling- Pain	Excision	-
156	Imaizumi [101]	2014	Japan	Male	7	birth	Left parotid gland	10 × 8	Enlargement- Pain- Dyspnea- Dysphagia	Medication Therapy	-
157	Kayhan [102]	2014	Turkey	Female	7	birth	Tongue	-	Carries- Occlusion problems	-	-
158				Female	12	3	Tongue	-	Swelling	Follow up	-
159				Female	29	11	Tongue	-	Enlargement- Pain- Bleeding	Follow up	-
160				Male	10	birth	Tongue	-	Gingival problems	-	-
161	Maboudi [103]	2014	Iran	Male	17	-	Gingiva	-	Enlargement- Bleeding	-	no
162	Neri [104]	2014	Italy	Female	11	birth	Right buccal mucosa- Upper lip	-	Swelling- Erythema- Fever	Antibiotic therapy-	-
163	Tiwari [105]	2014	India	-	Newborn	birth	Tongue		Swelling- Tongue protrusion- Difficulty with breathing	Surgical Excision	no
164	Usha [106]	2014	India	Female	13	1 month	Tongue	2 × 3	Sense of Burning- Occlusion impairment	-	-
165	Yoganna [107]	2014	India	Male	14	9	Left buccal mucosa	3 × 4	Swelling	Excision	no
166	De Souza [108]	2015	-	Male	13	11	Tongue	0.5	Blister Existence	-	no
167	Anpuram [109]	2015	India	-	New born	birth	submandibular region- floor of mouth- tongue	4 × 4.5	Mass existence	Excision	-
168	Bhayya [5]	2015	India	Male	8	6	Tongue	4 × 3	Enlargement- Difficulty with speech- Bleeding- Dysphagia	Excision	-
169	de Carvalho [110]	2015	Brazil	Male	6	-	Lower lip	0.3	Mass existence	Excision	-
170				Female	4	-	Lower lip	1	Mass existence- Difficulty with eating- Difficulty with speaking	Excision	-
171	Ganigara [111]	2015	India	Male	3	birth	Tongue	-	Enlargement- Difficulty with Occlusion- Dysphagia- Difficulty with Speaking	Sclerotherapy	-
172	Gupta [112]	2015	India	Female	2	birth	Tongue	-	Swelling- Protrusion	Excision	-
173	Kaur [113]	2015	India	Male	17	4	Right buccal mucosa	-	Swelling	-	-
174	Lee [114]	2015	Korea	Female	1	birth	Floor of mouth- Tongue	5 × 4.5	Swelling	Excision	-
175	Nagpal [115]	2015	India	Male	15	7	Tongue	-	Enlargement- Protrusion	Partial Glossectomy	no
176	Pammar [116]	2015	India	Female	45	2	Left buccal mucosa	0.5 × 1	Swelling	Excision	no
177	Beech [117]	2016	UK	Female	5	-	Tongue	-	Swelling- Pain	Follow up	-
178	Erugula [118]	2016	India	Female	16	3	Tongue	-	Swelling	Laser therapy	no

179	Gaddikeri [119]	2016	India	Male	70	-	Tongue	1 × 1	Enlargement-Difficulty with Speech-Bleeding-Dysphagia	Excision	-
180	Iliadou [120]	2016	Greece	Male	9	9	Right Tonsil	1.8 × 1.2 × 1.2	Mass feeling in throat	Bilateral tonsillectomy	-
181	Mechery [121]	2016	India	Female	20	19	Upper lip	3 × 1.5	Ulcers	Laser Therapy	no
182	Singh [121]	2016	India	Male	45	45	Alveolar ridge	-	Bleeding	Excision	no
183	Ugurlu [122]	2016	Turkey	Male	51	-	Alveolar ridge	3 × 2	Discoloration	Excision	no
184	Leao [123]	2017	-	Male	9	7	Tongue	2 × 1.5	Anesthetic complaints	Surgical Excision	no
185	Eren [124]	2017	Turkey	Male	26	-	Tongue	1.5 × 1.5	Mass feeling	Surgical excision	-
186	Gupta [125]	2017	India	Male	5	birth	Tongue	3 × 3	Mass Existence-Enlargement	Excision	-
187	Hwang [6]	2017	Korea	Female	10	6	Tongue	7 × 3.5 × 2	Swelling-Bleeding	Medication Therapy	no
188	Fernandes [126]	2018	-	Female	68	62	Lower lip	-	Mass Existence	Follow up	no
189	Puri [127]	2018	-	Female	23	-	Parotid region	-	Enlargement	Surgical excision	-
190	Leonel [128]	2017	-	Male	17	7	Tongue	3	-	Surgical excision	-
191	Alrashdan [1]	2018	Jordan	Male	42	40	Tongue	1.2	Mass Feeling	Excision	-
192	Bajpai [129]	2018	India	Female	12	birth	Tongue	-	Swelling	-	-
193	Bajpai [130]	2018	India	Male	51	1	Right parotid gland	3 × 3	Swelling	Surgical enucleation	-
194	Chinnakkulam [131]	2018	India	Female	50	48	Right parotid gland	5 × 6	Swelling- Pain	Surgical excision	-
195	Sharma [132]	2018	India	Male	15	5	Tongue	-	Mass existence-Sense of burning-bleeding	Sclerotherapy	-
196	Heidenreich [133]	2019	Brazil	Female	24	-	Tongue	-	Enlargement-difficulty with mastication and speech	Partial glossectomy	-
197	Abdul Wadood [134]	2019	Iraq	Female	22	-	Soft palate	-	Bleeding	Hydrocortisone sodium succinate injection	no
198	Biher [135]	2019	India	Male	41	-	Tongue	-	-	Surgical excision	-
199	Farronato [136]	2019	Italy	Male	4	-	Tongue	-	Esthetic problems-Dysphagia-difficulty with speaking-airway obstruction-Deformity	Resection	-
200	Kim [137]	2019	Korea	Male	7	birth	Tongue	-	Enlargement	Partial glossectomy	-
201	Matharu [8]	2019	Uk	Female	4	birth	Lower lip	0.55 × 0.17 × 0.7	Mass Existence	Cryotherapy	-
202	Nelson [7]	2019	Malaysia	Female	24	4	Tongue	-	Pain in tooth	Excision	-
203	Yalcin [3]	2019	Turkey	Female	15	-	Left buccal and retro molar mucosa	-	Blisters	Excision	no
204	De Oliveira [138]	2020	-	Male	9	birth	Tongue	3	Mass feeling-Pain	Laser Therapy	no
205	Hass [139]	2020	-	-	-	-	Tongue	0.5	-	Excision	-
206	Aires [140]	2020	Brazil	Male	9	birth	Tongue	-	Bleeding-Pain-Problem with chewing	Laser therapy	no
207	Shah [141]	2020	India	Female	10	birth	tongue	-	Swelling	Surgical Excision	-
208	Jha [142]	2020	India	-	8	6	Right side of neck adjacent to ramus	4.7 × 2.8	Swelling	Surgical excision	no
209				Female	26	24	Right submandibular gland	4.7 × 4.6	Swelling	Surgical Excision	no
210	Recent Case	2020	Iran	Female	50	48	Left buccal mucosa	1 × 1.5	Mass existence	-	-

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