Lymphangioma Of Tongue

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Citation

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Abstract

Lymphangiomas are uncommon congenital tumors of the lymphatic system, which usually occur in the head and neck but involvement of the tongue is rare. We report the case of a 2-year-old child with a lymphangioma of the tongue. Early recognition and surgical excision of these tumors are essential for an optimal functional outcome. The histopathology and treatment of tongue lymphangioma are reviewed.

CASE REPORT

A 2-year-old male child presented to ENT OPD of Lok Nayak hospital with enlargement of tongue for last 1 year. It was insidious in onset with history of difficulty in eating solids. There was no history of respiratory difficulty, trauma, pain, bleeding or sudden increase in the size of swelling. Examination revealed diffusely enlarged tongue, protruding and keeping the mouth permanently open. A small area of ulceration over the dorsum of tongue on anterior part was also present. On palpation it was soft to firm in consistency. FNAC of the swelling yielded only blood, hence a clinical diagnosis of hemangioma was made.

The child was taken up for V glossoplasty. A preoperative tracheostomy was done. The anterior half of the tongue in inverted V shaped was removed. The tumor was having large cystic spaces filled with lymph like fluid. The part of tumor going into the base tongue was left and injected with sclerosing agent ie 25% Dextrose. It was then sutured in midline to give tongue it normal shape. Bilateral submandibular ducts were identified and cannulated. Histopathological diagnosis was lymphangioma. The tracheostomy tube was removed after 48 hours and the postoperative period was uneventful.

Figure 1

Figure 1: clinical photograph showing macroglossia

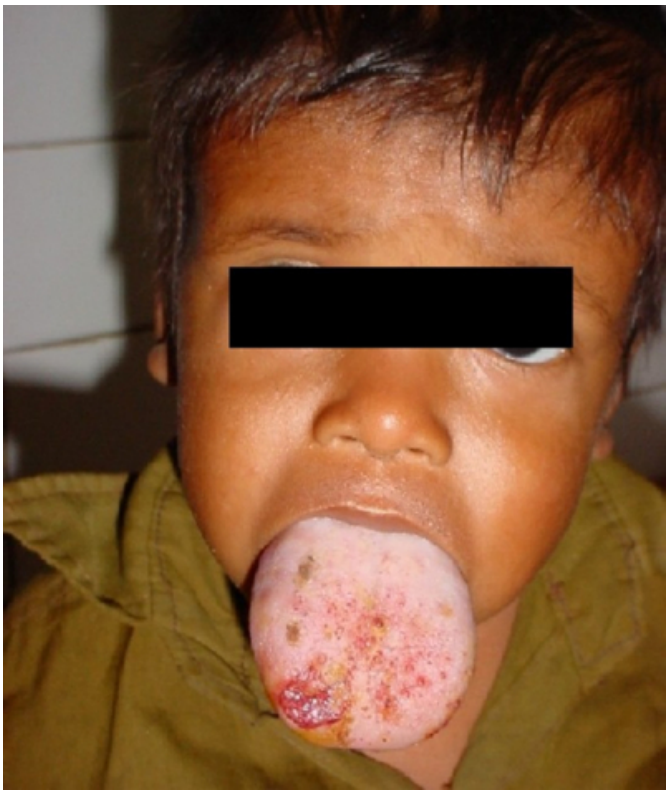
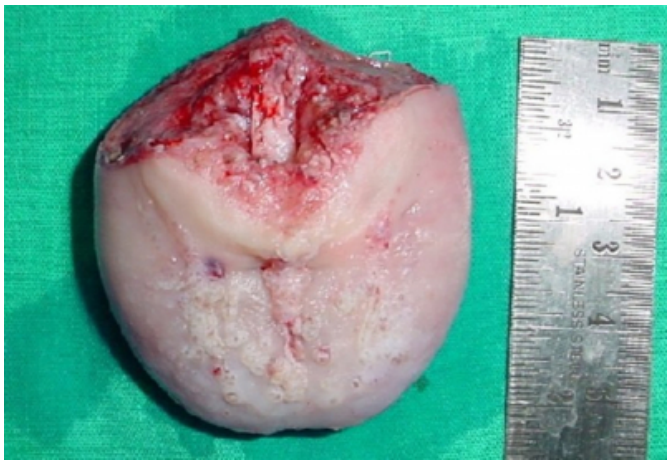


Figure 2

Figure 2: excised tongue specimen



DISCUSSION

Virchow (1854) gave the first accurate description of lymphangioma. The Lymphangiomas are localized in the head and neck area in about 75% of cases and about 80% of these cases are children less than 2 years old[1]. Although tongue is a rare site for lymphangioma it is the most common cause for macroglossia [2]. In the oral cavity, lymphangioma is a rare, non-odontogenic, benign neoplasm, which originates from lymph vessels. This lesion is common in the first decade of life and mostly occurs on the dorsal surface and lateral border of the tongue and rarely arises on the palate, gingiva, buccal mucosa and lips. These tumors rarely regress and they keep on growing slowly with each episode of upper respiratory tract infection because of deposition of fibrous tissue around dilated lymphatic channels [3]. A rapid increase in size is seen in infection or bleeding, when they can cause speech difficulty respiratory distress, dysphagia and sleep apnea[4]. Complete surgical excision is treatment of choice but this is not always possible for example extensions into base tongue, floor of mouth, larynx, neurovascular structures of neck and mediastinal extension. Recurrent, residual, unresectable or surgically challenging tumors are treated with intralesional injection of sclerosing agents like 25% dextrose, hypertonic saline, bleomycin, aethoxysklerol, OK-432 (picibanil) [5] before surgery. A change in consistency of the tumor, manifests by softening, is followed by marked shrinkage. Satisfactory results can be obtained, resulting in definite reduction in size and improvement in cosmetic appearance. In recent years Carbon dioxide and Neodymium Yitrium Aluminum Garnet (Nd-YAG) laser photocoagulation surgery has become popular. Earlier steroids, electrocoagulation, cryotherapy or radiation therapy has been used with variable results but most effective treatment is surgery [6].

On histopathological examination a complex vascular malformation of the lymphangioma-hemangioma type, involving extensively the deep musculature of the tongue and extension into base of tongue, in background of variable muscle degeneration and marked fibrosis was found. The lymphangioma consists of multiple, intertwining lymph vessels in a loose fibrovascular stroma, sometimes with scattered aggregates of lymphoid tissue. The lymphatic vessels of a lymphangioma are lined by a single layer of endothelial cells with flattened, occasionally plump, nuclei. The vessels may be capillary in size, with a very attenuated lumen or may be so dilated that the cystic areas can be visualized at surgery. Oral lesions are more likely to contain the dilated vessels. The vessels are filled with a uniform, eosinophilic, proteinaceous fluid with occasional erythrocytes and leukocytes.

There is no encapsulation of lymphatic vessels, even with the tumors, which appear well circumscribed clinically. Deeper lesions show vessels interspersed between adipocytes and striated muscle bundles. Deeper lesions also tend toward greater vessel dilation.

Lymphangioma can be classified into four categories:

1. lymphangioma simplex (capillary lymphangioma, lymphangioma circumscriptum), composed of small, thin-walled lymphatics
2. cavernous lymphangioma, comprised of dilated lymphatic vessels with surrounding adventitia
3. cystic lymphangioma (cystic hygroma), consisting of huge, macroscopic lymphatic spaces with surrounding fibrovascular tissues and smooth muscle
4. benign lymphangioendothelioma (acquired progressive lymphangioma), lymphatic channels appear to be dissecting through dense collagenic bundles.

These categories are somewhat artificial and many lesions are combinations of all categories. Occasional lesions demonstrate proliferation of lymphatic channels with another connective tissue component, primarily smooth muscle cells (lymphangiomyoma).

The lymphatic endothelial cells in these or more routine

lymphangiomas can be identified via positive immunoreactivity with factor VIII-associated antigen and CD31 [7]. Immunohistochemistry is not a particularly reliable means of distinguishing lymphangioma from hemangioma, but this is not usually a problem because of the large number of erythrocytes in the vessels of the latter.

CONCLUSION

Because of the nonencapsulated and “infiltrating” nature of the lymphangioma of tongue, complete removal is invariably impossible and inadvisable. Surgical debulking of the tumor is, therefore the treatment of choice with objective of having a functional tongue. It should also be understood that additional procedures might be required as the child grows. For small recurrent and unresectable tumors sclerotherapy is useful. A preoperative tracheostomy is always better in cases of large tongue hemangioma.

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