DECEPTIVE ORAL LESION IN A CHILD:

A RARE CASE REPORT OF TONGUE LYMPHANGIOMA

ABSTRACT : Lymphangiomas are rare, benign developmental malformations of the lymphatic system, resulting from sequestration or abnormal proliferation of lymphatic vessels. They most commonly occur in the head and neck region, with the tongue being one of the frequent intraoral sites. A 5-year-old girl was referred to the Pedodontics OPD with a reddish, irregular lesion on the back of her tongue, present since 7 months of age, but without a proper diagnosis. The only symptom was intolerance to spicy food. Clinically, the lesion mimicked posterior midline atrophic candidiasis. On examination, the lesion was soft, non-tender, and localized without significant enlargement. No history of trauma, systemic illness, or similar lesions elsewhere was noted. An Incisional Biopsy was performed from the borders of the lesion, and histopathological examination revealed features consistent with Lymphangioma.This case report highlights the vital role of the Pedodontist in diagnosing rare tongue lesions in children through careful clinical evaluation and biopsy.

KEYWORDS: candidiasis, posterior aspect of tongue, incisional biopsy, lymphangioma

INTRODUCTION

Lymphangiomas are non-cancerous malformations of lymphatic tissue that develop when portions of lymphatic tissue become sequestered during development. These lesions most commonly appear in the head and neck area, accounting for roughly 50–70% of cases. Around half of lymphangiomas are present at birth, and nearly 90% become apparent by the age of two. Tiny lymphangiomas under 1 cm in size can be found on the alveolar ridge, and in such cases there is approximately a 2:1 ratio of males to females.[1]This slow-growing, painless soft-tissue mass was first described in 1828 by Redenbacher and then in 1854, Virchow first described lymphangiomas of the tongue. They are benign developmental malformations rather than true neoplasms characterized by abnormal proliferation of lymphatic channels. According to the another school of thought, they may originate from endothelial fibrillar membranes which bulges from the cystic wall and penetrates the surrounding tissue which canalize and form more cyst.[2]

Lymphangiomas are divided into three types histologically :

1. Simple (capillary) – made of small, thin-walled lymphatic vessels,
2. Cavernous – composed of larger, dilated lymphatic channels, and
3. Cystic (cystic hygroma) – featuring large, macroscopic cystic spaces.[2]

Despite the fact that lymphangiomas are rare lesions, they contribute to 4% of all vascular tumours and 25% of vascular tumours in children.The tongue is the most commonly affected site in the oral cavity, and sites such as the palate, gingiva, and alveolar ridge of the mandibleare rarely affected. Lymphangiomas usually manifest as papillary lesions with the same colour as the adjacent mucosa.

Deep lymphangioma lesions show up as soft, spread-out lumps that feel soft to the touch and have a colour similar to the surrounding normal tissue rather than looking very different.Occasionally, oral lymphangiomas are associated with syndromes like Turner's Syndrome, Noonan's Syndrome, Trisomies, Cardiac Anomalies, Foetal Hydrops, Foetal Alcohol Syndrome, And Familial Pterygium Colli. Tongue lymphangiomas are usually superficial with a pebbly surface resembling a cluster of translucent vesicles.The anterior two-thirds of the tongue is the most commonly affected site leading to swelling and enlargement of the tongue . Patients with tongue lymphangioma usually suffer from speech disturbances, poor oral hygiene, and bleeding from the tongue when exposed to trauma .

The etiology of lymphangioma remains unclear, but different hypotheses have been suggested to account for the pathogenesis of lymphangioma. One of the major theories proposes that thelymphatic system originates from five poorly developed sacs arising from the venous system. In the maxillofacial region, an endothelial outpouching from the jugular sac spread to form the lymphatic system. Another hypothesis concludes that the lymphatic system originates from mesenchymal clefts in the venous plexus reticulum and spread centripetally toward the jugular sac.[3]

In Pediatric patients, thorough clinical evaluation by a Pedodontist is essential for formulating a comprehensive differential diagnosis and identifying atypical characteristics that warrant further investigation. This case report emphasizes the pivotal role of a Pedodontist in the accurate and timely diagnosis of an oral lesion that ultimately proved to be a lymphangioma.

CASE REPORT

A 5-year-old female was referred to the Department of Pediatric and Preventive Dentistry for evaluation of a persistent lesion on the posterior dorsum of the tongue that had been present since approximately 7 months of age, with no prior definitive diagnosis. The primary symptom reported was intolerance to spicy food, manifesting as a burning sensation with no history of trauma, systemic illness, or similar lesions elsewhere.(Figure 1a).

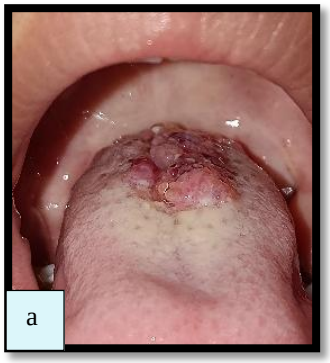

Figure 1: (a)Irregular elevated reddish lesion on posterior dorsum of the tongue

Intraoral examination revealed an irregular, elevated, reddish lesion located on the midline of the posterior region of tongue; the lesion was soft, cystic, and non-tender, with a pebblyappearence. Additionally, white, scrapable patches were observed on the surrounding mucosa. Dental examination also showed carious lesions involving the mandibular primary second molars.(Figure 2a). All relevant clinical findings and history were documented in chronological order to aid in differential diagnosis and further management.

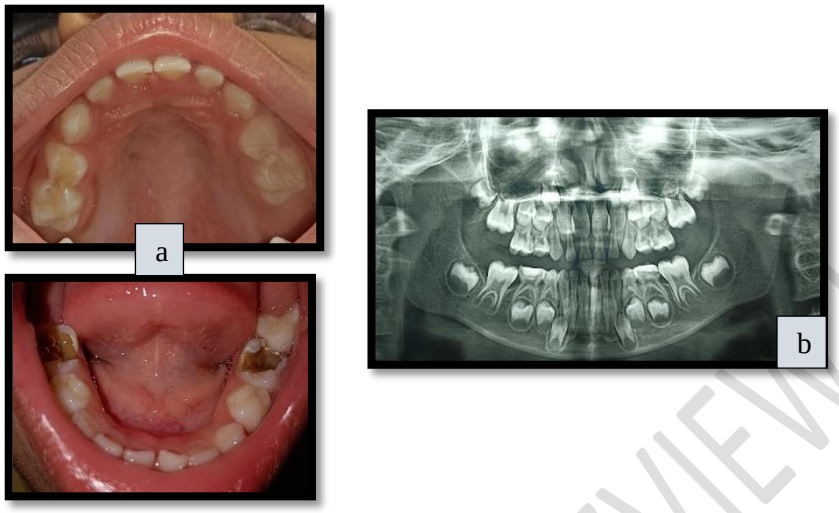

Figure 2: (a) Intraoral view of dentition showing dentinal caries of mandibular primary second molars , (b) OPG

A provisional clinical diagnosis of a posterior midline atrophic candidiasis variant was made based on the lesion's appearance and chronicity. Differential diagnoses considered included geographic tongue, lingual thyroid, erythroplakia, squamous cell carcinoma, granular cell tumor, lymphangioma, and various nutritional deficiency-related glossitis, reflecting the broad spectrum of red and irregular tongue lesions that may mimic candidal involvement in clinical practice. To further evaluate the condition and exclude other potential etiologies, a comprehensive diagnostic workup was undertaken. Routine blood investigations were performed to assess systemic status and rule out hematologic abnormalities.

An ultrasound examination of the neck ruled out the presence of ectopic thyroid tissue. Histopathological confirmation was advised through an incisional biopsy to establish a definitive diagnosis.

HISTOPATHOLOGY REPORT

Histopathological examination of the incisional biopsy specimen, which included three soft tissue fragments (measuring approximately 0.6 × 0.5 × 0.5 cm, 0.4 × 0.3 × 0.3 cm, and 0.4 × 0.4 × 0.4 cm), revealed hyperplastic parakeratinized stratified squamous epithelium overlying a delicate, collagenous connective tissue stroma. Within the superficial epithelial layer, eosinophilic filamentous structures morphologically consistent with candidal hyphae were identified, along with scattered hematoxyphilic granular material that suggested necrotic debris. The underlying connective tissue was notably cellular and contained multiple

sinusoidal-like spaces with mucin-like eosinophilic areas and occasional extravasated red blood cells.

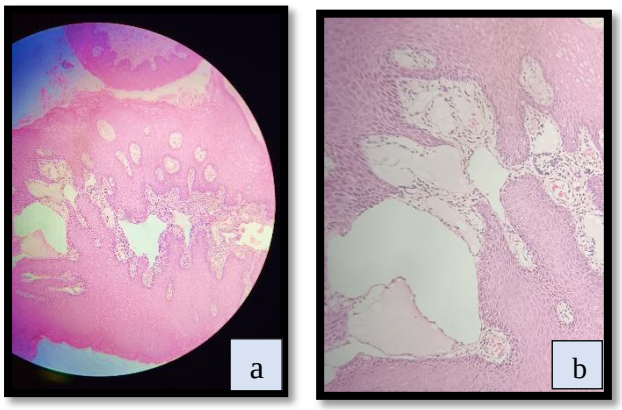

Figure 3 : (a) Histopathology image

(b)multiple dilated lymphatic-type sinusoidal spaces within a collagenous connective tissue stroma characteristic of lymphangioma.

Numerous dilated, lymphatic-type spaces were also evident within the papillary connective tissue, consistent with the characteristic appearance of lymphatic malformation. These histological features supported a definitive diagnosis of lymphangioma, a benign lymphatic malformation marked by proliferating lymphatic channels within a fibrovascular stroma, with superadded candidal colonization of the overlying epithelium.(Figure 3a,3b).

With the definitive diagnosis of lymphangioma with superadded candidiasis established, a comprehensive treatment plan was formulated. The patient was referred back to the Department of Pediatric Surgery for specialized management of the lymphangioma, and topical antifungal therapy in the form of a candid mouth paint was prescribed to alleviate the symptoms associated with superadded candidal infection.
Additionally, propranolol therapy was advised by the Department of Pediatrics as a potential systemic treatment to reduce the lymphangioma lesion under careful clinical monitoring, in conjunction with ongoing follow-up examinations.Multidisciplinary follow-up aims to assess both the response of the lymphatic malformation to therapy and the overall oral health status, ensuring timely intervention for any evolving clinical needs.

DICSUSSION

Lymphangioma is a benign congenital anomaly of lymphatic vessels rather than a true tumor. On the tongue, it often appears superficially as a cluster of small, translucent, pebbly vesicles that look like frog eggs or tapioca pudding. Deeper lesions feel like soft, poorly defined masses and do not show the typical surface pattern.[4,5]Lesions that might be misdiagnosed for lymphangioma include Haemangioma, Congenital Hypothyroidism, Amyloidosis, Neurofibromatosis and Primary muscular hypertrophy.[3]

Lymphangiomas treatment depends on their size, location and infiltration to the surrounding tissues. Surgical excision is the treatment of choice with the inclusion of a surrounding border of normal tissue. Recurrence rate is around 39% because of its infiltrative nature and surgeons often worry in achieving complete resection.[2]

Various treatment options have alsobeen examined, including theinjection ofsteroids orsclerosing agents, electrocoagulation, cryotherapy andlaser surgery.Intralesional steroids cause a significant increase in mast cell density, reduced transcription of cytokines,decreased platelet-derived growth factor A and B, and decreased basic fibroblast growth factor. These mechanisms alter cellular functions, resulting in regression of the lymphatic malformation without a significant inflammation reaction.

Sclerosing agents such asbleomycin, OK-432, sodium tetradecyl sulfate, cyclophosphamide, andhypertonic saline arefrequently utilized inthetreatment oflymphangiomas. There isextensive literature onthe useofbleomycin fortreating congenital lymphatic anomalies inthe head andneck. Itexerts asclerosing effect ontheendothelial lining of blood vessels.Benefits ofLaser therapy inthetreatment oflymphangioma include reduced bleeding andfaster healing. Limitationlike effectiveness ondeep infiltrated lesions, potential scarring and more pain and discomfort postoperatively makes themain drawbacks.[6]The Nd:YAG Laser appears to be one of the safest therapeutic options rarely proposed to treat oral cavity lesions.[7]Blockade of VEGF by bevacizumab is used as an effective treatment for capillary hemangioma and diabetic retinopathy and has been proved to be a treatment for lymphatic malformation.[8]

Propranolol is believed to exert its effect by down-regulating the Raf/mitogen-activated protein kinase signaling pathway, leading to decreased expression of vascular endothelial growth factor (VEGF). This mechanism was supported by observations in a patient with diffuse lymphangiomatosis, in whom elevated plasma VEGF levels before treatment significantly declined following successful propranolol therapy, suggesting that propranolol may inhibit lymphangiogenesis and reduce lymphatic malformation growth through suppression of VEGF-mediated signaling.[9]

CONCLUSION

Although lymphangioma is a benign congenital lymphatic malformation and its occurrence in the tongue is exceptionally rare, cliniciansincluding dental and other healthcare professionalsshould maintain a high index of suspicion for such lesions to facilitate early recognition and accurate diagnosis. Early identification enables appropriate management, which is critical to preventing potentially severe complications; if traumatized or secondarily infected, these lesions can enlarge and compromise the upper airway, posing a risk of life-threatening obstruction in the absence of prompt intervention..[3]

In this case, the patient's sole complaint was a localized burning sensation, with no evidence of impairment in speech, deglutition, or swallowing. As the symptom burden was minimal and there were no functional deficits, conservative management was undertaken. The treatment plan began with oral propranolol administered under the supervision of a pediatrician, accompanied by regular clinical evaluations to monitor the lesion's response. The Pediatric dentist played a pivotal role, as the lesion had previously been undiagnosed despite earlier assessments; the specialist identified the abnormal soft tissue presentation, initiated histopathological confirmation, and thereby facilitated development of an

appropriate interdisciplinary management strategy.This case underscores the importance of comprehensive intraoral examination and Pediatric dental evaluation for identifying atypical mucosal lesions.

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