Oral Lymphangiomas: A Rare Presentation of Two Cases with Dermoscopic Findings

Abstract

Lymphangiomas are benign hamartomatous malformations, infrequently seen in the oral cavity. They can cause aesthetic issues and functional impairments if not treated in the initial stages, thus necessitating the need to diagnose early. Two presentations were observed: a 35-year-old male with classical frog-egg appearance involving lower buccal mucosa with macrocheilia for two years, which was diagnostic on mucoscopy and histopathology, confirming acquired capillary lymphatic malformation. The second case was of a 12-year-old girl with a gradual appearance of reddish lesions over the tongue and episodic history of bleeding, which was confirmed on mucoscopy, and histopathology is highlighted. Mucoscopy of both the cases showed yellowish-pink lacunae with a hypopyon-like feature and surrounding pale septa. We present two cases of oral lymphangiomas with different presentations and highlight the importance of dermoscopy of oral lesions to bypass invasive techniques such as biopsy.

Keywords: *Acquired, buccal mucosa, hypopyon, lacunae, mucoscopy, oral lymphangiomas, pale septa, tongue*

Jayati Shailesh Dave, Bhushan Telhure, Sunanda Arun Mahajan, Meghana Rane

Department of Dermatology, Seth G.S. Medical College and K.E.M Hospital, Mumbai, Maharashtra, India

Introduction

Lymphangiomas, first described by Redenbacher in 1828, are benign, hamartomatous malformations arising from sequestrated lymphatic tissue.[1] This sequestrated tissue cannot develop adequate anastomosis with larger lymphatic channels. It thus presents as localized areas of lymphatic blockage. Fifty percent of them present at birth, and 90% of them usually present by the age of two years. Acquired lymphangiomas occur secondary to damage to previously normal lymphatics. Lymphangiomas have a marked predilection for the head and neck regions, which accounts for about 50–70%.[2] They are uncommon in the oral cavity. We report two cases of intraoral lymphangiomas with different presentations and one of them being acquired in nature.

Case report 1

A 35-year-old man presented with complaints of asymptomatic swelling of the lower lip with lesions over the lower buccal mucosa for two years [Figure 1a]. He did not have a history of any systemic illness or any treatment course in the past. Cutaneous examination revealed multiple, clear fluid-filled vesicles with closely clustered pink papilliform lesions over the lower and left buccal mucosa [Figure 1b]. We kept differential diagnoses as mucosal warts, acquired lymphangioma, and oral florid papillomatosis and decided to perform a biopsy. Dermoscopy (polarized) showed multiple, yellowish-pink, well-circumscribed round areas known as lacunae which were surrounded by pale septa on a yellowish background. Few short linear vessels and red dots were also noted [Figure 2a]. These lacunae showed a hypopyon-like feature [Figure 2b]. Histopathological evaluation revealed acanthosis and hyperkeratosis of the epidermis. Within the papillary and reticular dermis, there were multiple endothelial cells lined with dilated lymphatic channels with eosinophilic lymph fluid, confirming the diagnosis of acquired lymphangioma [Figure 4a]. The patient was started on cryotherapy sessions with little improvement.

Case report 2

A 12-year-old girl presented to the outpatient department with lesions over the

How to cite this article: Dave JS, Telhure B, Mahajan SA, Rane M. Oral lymphangiomas: A rare presentation of two cases with dermoscopic findings. Indian Dermatol Online J 2023;14:80-2.

Received: 11-Mar-2022. **Revised:** 30-May-2022. **Accepted:** 01-Jun-2022. **Published:** 14-Dec-2022.

Address for correspondence:
Dr. Sunanda Arun Mahajan, Department of Dermatology, Seth G.S. Medical College and K.E.M Hospital, Mumbai, Maharashtra, India. E-mail: sunandamahajan@gmail.com

Access this article online

Website: www.idoj.in

DOI: 10.4103/idoj.idoj_154_22

Quick Response Code:

tongue for two years which were insidious in onset and gradually progressive in size and number. She had a history of episodic bleeding from the lesions after trivial trauma. She had achieved age-appropriate growth and puberty. On cutaneous examination, we found multiple, soft, 0.5mm discrete, erythematous to violaceous, non-tender papules over the dorsum of the tongue. Few ill-defined pink-colored verrucous papules were also present over the dorsum of the tongue [Figure 3a]. We kept angiokeratoma circumscriptum, venous malformation, hemangioma, and hemolymphangioma as other differentials. Dermoscopy (polarized) was suggestive of multiple pale-yellow papilliform projections with yellowish-white and blood-filled red lacunae surrounded by pale septa on a yellow background. Few lacunae showed a hypopyon-like feature [Figure 3b]. Histopathology of the cut section of the verrucous lesion and soft papule showed acanthosis and hyperkeratosis of the epidermis. Within the papillary and reticular dermis, there were multiple dilated endothelium-lined lymphatic channels with many red blood cells (RBCs) suggestive of hemolymphangioma [Figure 4b]. The patient was referred to a plastic surgeon and otolaryngologist for further treatment.

Discussion

Lymphangiomas of the oral cavity are most commonly seen on the dorsum of the tongue, especially on the anterior two-thirds presenting as macroglossia sometimes. The less commonly involved sites include the soft palate, buccal mucosa, gingiva, floor of the mouth, and lips.[1] There are three types: (1) capillary lymphangioma simplex/superficial microcystic with cavities less than 2 cm^3 presenting as pink/flesh-colored clustered papules/vesicles resembling frog spawn, (2) cavernous lymphangioma/macrocystic with cavities more than 2 cm^3 presenting as subcutaneous rubbery nodules without surface/texture change, (3) cystic hygroma that are cystic and larger than cavernous lymphangiomas, commonly occurring in the head and neck regions, and (4) mixed variant having both microcystic and macrocystic components.[1,2]

Acquired lymphangiomas have been reported secondary to insult to previously normal lymphatics. Insults such as radiation therapy, denture-induced trauma, tuberculous adenitis, scrofuloderma, and intra-abdominal and pelvic surgery have been reported.[3]

Clinically superficial intraoral lymphangiomas may present as pebbly lesions with groups of translucent vesicles which appear like tapioca pudding or frog eggs. They can also present as red or blue papules/nodules, which is due to rupture of the blood capillary into the lymphatic space. Deeper lesions appear as soft ill-defined masses.[1,2]

The diagnosis of intraoral lymphangiomas is usually clinical, but sometimes it can create a diagnostic dilemma. Mucoscopy and histopathological evaluation are diagnostic

Figure 1: (a) Diffuse swelling of the lower lip, white papules over the right side of the buccal mucosa with scaling over the outer aspect of the lower lip. (b) Multiple clear fluid-filled vesicles with closely clustered pink papilliform lesions over the lower buccal mucosa mimicking a cluster of frog eggs

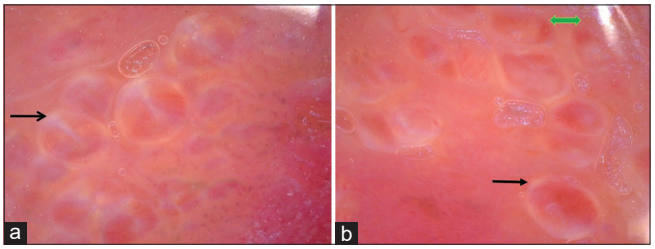

Figure 2: (a) Dermoscopy (polarized, 20×) showed a yellowish background studded with multiple yellowish-pink well-circumscribed roundish areas known as lacunae which were surrounded by pale septa (black arrow). Few short linear vessels and red dots are seen in the background. (b) Dermoscopy (polarized, 20×) demonstrates pale septa (black arrow); sedimentation of blood to the bottom of the lacuna gives a hypopyon-like/half-and-half lacuna appearance (green arrow)

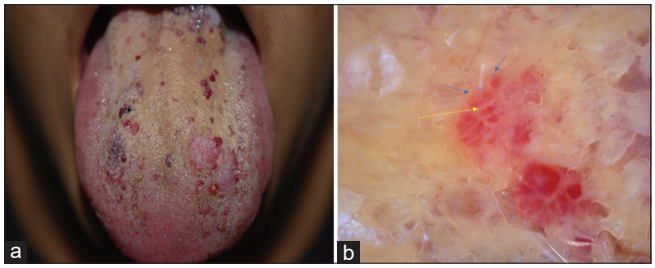

Figure 3: (a) Multiple soft 0.5mm discrete erythematous to violaceous, non-tender papules over the dorsum of the tongue. Few ill-defined pink-colored verrucous papules were also present over the dorsa of the tongue. (b) Dermoscopy (polarized, 10×) was suggestive of a yellowish background and multiple pale-yellow papilliform projections with yellowish-white and blood-filled red lacunae (blue arrow) surrounded by pale septa (yellow arrow). Few lacunae show a hypopyon-like feature

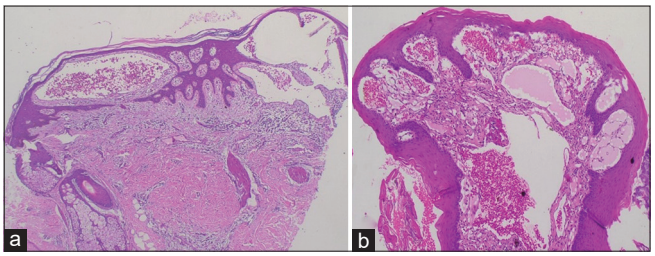

Figure 4: (a) Histopathological examination (H&E,10X magnification) of lip lesion revealed acanthosis and hyperkeratosis of the epidermis. Within the papillary and reticular dermis, there were dilated lymphatic channels lined by endothelial cells. (b) Histopathological examination (H&E,10X magnification) of tongue lesion showed acanthosis and hyperkeratosis of the epidermis. Within the papillary and reticular dermis, there were dilated lymphatic channels filled with eosinophilic lymph fluid and multiple RBCs

tools which can come to the rescue. Like cutaneous lymphangiomas, mucoscopy of oral lymphangiomas presents with yellow lacunae surrounded by pale white septa. The lacunae appear yellow due to the inclusion of lymph fluid, whereas the reddish-/bluish-colored appearance may be due to blood mixed with lymph. Different amounts of blood in lacunae may cause various dermoscopic features such as focal reddish areas, pink diffuse coloration, and reddish-to-violaceous lacunar structures.[4] The blood in lacunae accumulates in the lower part, whereas lymph in the upper part, giving an appearance of the hypopyon sign.[4,5] Gencoglan *et al*.[4] have proposed that as sedimentation of blood occurs, corpuscles aggregate as per their density with cellular components lying at the bottom and serum at the upper part, causing a color transition effect from dark to light in lacunae like the hypopyon seen in Sneddon–Wilkinson disease.[5] Jha *et al*. have described this hypopyon in metaphoric terminology as half-and-half-lacunae.[5] Histologically, lymphangiomas consist of solitary or grouped dilated lymphatic channels lined by endothelial cells in the papillary dermis. These dilated cystic channels contain an eosinophilic material and sometimes the RBCs. Imaging modalities such as color Doppler and computed tomography angiogram/magnetic resonance imaging can help identify the depth of involvement and coexisting vascular involvement, if any.[1]

Dermoscopic differentials of oral lymphangiomas include hemangiomas, angiokeratomas, molluscum contagiosum, and warts. The presence of hypopyon-like features and clear fluid in the lacunae can be helpful to distinguish lymphangiomas from hemangiomas.[4] On dermoscopy, angiokeratomas are characterized as multiple reddish-blue lacunae, whitish veil, and milky-red areas corresponding with dilated and congested vessels and hyperplastic squamous epithelium.[6] Warts present as white structureless areas with white projections.[6] Molluscum contagiosum has a typical dermoscopic feature presenting with polylobular whitish-yellow amorphous structure with surrounding crown vessels that do not cross the center.

Complications of oral lymphangioma include cosmetic deformity, macroglossia, difficulty in mastication, impaired speech, and secondary bacterial infections, and larger ones can cause airway obstruction.

Treatment options for lymphangiomas include surgical excision, cryotherapy, electrocautery, sclerotherapy, embolization, and laser therapy.[1]

Intraoral lymphangiomas, although rarely encountered, can be easily diagnosed with clinical acumen, dermoscopy, and histopathology. We report two cases with different presentations and their dermoscopic findings, one of them being acquired in nature. Early diagnosis and prompt treatment can prevent aesthetic and functional complications associated with the malformation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgement

We thank Dr. Uday Khopkar for the histopathological examination.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Kurude AA, Phiske MM, Kolekar KK, Nayak CS. Lymphangiomas: Rare presentations in oral cavity and scrotum in pediatric age group. Indian J Dermatol Venereol Leprol 2020;86:230.
2. Sunil S, Gopakumar D, Sreenivasan BS. Oral lymphangioma-Case reports and review of literature. Contemp Clin Dent 2012;3:116-8.
3. Ghalamkarpour F, Asadi-kani Z, Moradi A, Zaresharifi S, Khazaei P. Acquired lymphangioma circumscriptum secondary to tuberculosis: A rare case report. Dermatol Ther 2020;33:e13463.
4. Gencoglan G, Inanir I, Ermertcan AT. Hypopyon-like features: New dermoscopic criteria in the differential diagnosis of cutaneous lymphangioma circumscriptum and haemangiomas. J Eur Acad Venereol 2012;26:1023-5.
5. Jha AK, Lallas A, Sonthalia S. Dermoscopy of cutaneous lymphangioma circumscriptum. Dermatol Pract Concept 2017;7:37-8.
6. Kumar Jha A, Vinay K, Sławińska M, Sonthalia S, Sobjanek M, Kamińska-Winciorek G, *et al*. Application of mucous membrane dermoscopy (mucoscopy) in diagnostics of benign oral lesions - literature review and preliminary observations from International Dermoscopy Society study. Dermatol Ther 2021;34:e14478.