Case Report

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Lymphangiectasia Mimicking Lichenoid Contact Reaction

Dentistry Section

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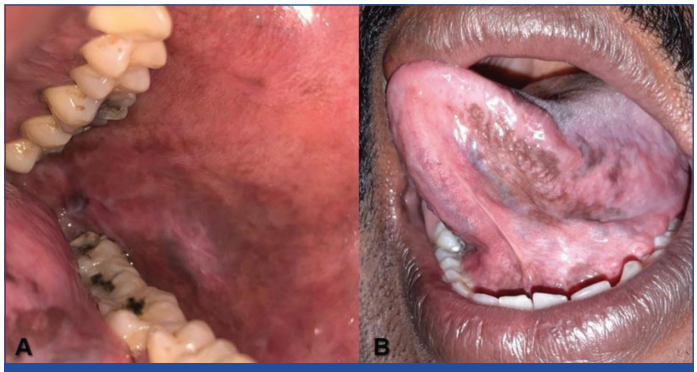
ABSTRACT

Lymphangiectasia, otherwise known as acquired lymphangioma, is unusual to occur in the oral cavity. Among the frequently recognised forms of congenital and acquired entities, the acquired form of the lymphangiectasia may be caused due to trauma or obstruction to the lymphatic system. Commonly, this condition is asymptomatic; however, the needs of aesthetics and functions warrant treatment. Hereby, authors present a case report of a 55-year-old male patient, who came with a chief complaint of burning sensation in his left cheek region for the past three weeks. Clinically, silver amalgam restoration was evident in relation to the tooth numbers 26,27,36 and 37. Soft tissue examination revealed evidence of greyish radiating striae interspersed with hyperpigmentation on the left buccal sulcus and left lateral border of the tongue. Considering the clinical features, a working diagnosis of lichenoid contact reaction was proposed. Further, an incisional biopsy was performed. Histopathological examination showed thin stratified squamous epithelium with vacuolar spaces, which strongly supported the diagnosis of lymphatic malformation. The patient was symptomatically managed with topical triamcinolone acetonide 0.1% twice daily for two weeks. At two weeks follow-up, the patient was symptom-free and hence, the medication was withdrawn. The patient is currently under regular follow-ups.

Keywords: Burning sensation, Greyish radiating striae, Hyperpigmentation, Silver amalgam restoration

CASE REPORT

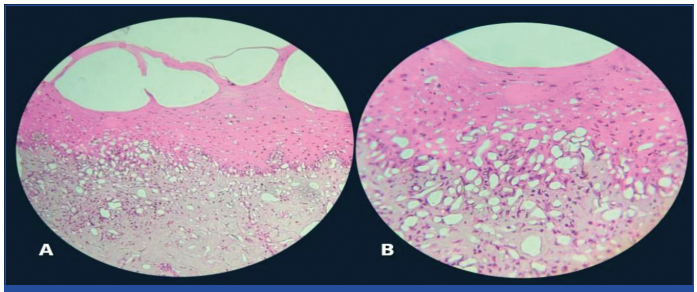
A 55-year-old male reported to the Department of Oral Medicine and Radiology with a chief complaint of burning sensation in the left inner cheek region for the past three weeks. Initially, burning sensation was only after intake of spicy food which gradually increased with time. Medical history was non contributory. On general examination, all vitals were normal. On local examination, intraorally, greyish radiating striae interspersed with hyperpigmentation was evident on the left buccal sulcus alongside 36, 37, 38 which was non scrapable and lesion was non tender [Table/Fig-1a]. Similar diffuse hyperpigmentation was evident on the left lateral border of ventral surface of the tongue sparing the tip [Table/Fig-1b]. Using Visual Analogue Scale (VAS) the burning pain score was denoted as 5 [1]. Contributing the soft tissue finding, silver amalgam restoration was evident in relation to the tooth numbers 26, 27, 36 and 37. On lymph node examination, no abnormality was detected.



[Table/Fig-1]: (a) Greyish radiating striae interspersed with hyperpigmentation on the left buccal sulcus; (b) Diffuse hyperpigmentation on the left lateral borders of ventral surface of the tongue sparing the tip.

Based on this clinical presentation, a provisional diagnosis of lichenoid contact reaction, either due to amalgam restoration or betel quid, was considered. Tobacco-induced hyperpigmentation, postinflammatory pigmentation and oral melanotic macule were considered as other differential diagnosis.

After obtaining the informed consent, routine haematological investigations were performed, which were within normal range and incisional biopsy was done in the left buccal mucosa. Histopathological examination revealed thin stratified squamous epithelium with vacuolar spaces and juxtaepithelium showed plenty of dilated endothelial lined lymphatic channels, deeper section showed the presence of dense fibrosis with few inflammatory cells [Table/Fig-2a,b]. Correlating the clinical and histopathological observation, a final diagnosis of lymphangiectasia involving the left lateral border of tongue and left buccal mucosa was arrived at.

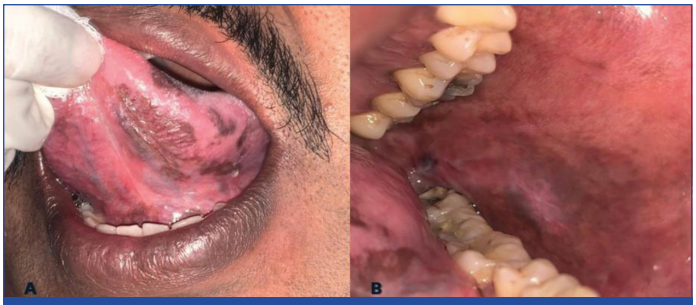


[Table/Fig-2]: (a) Photomicrograph displaying thin stratified squamous epithelium with vacuolar spaces (H&E, 4X); (b) Juxtaepithelium showed plenty of dilated endothelial lined lymphatic channels (H&E, 20X).

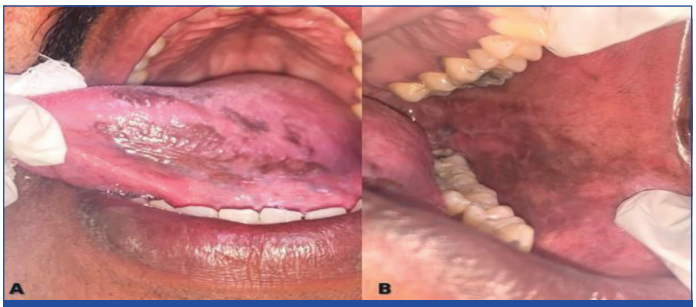
The patient was symptomatically treated with topical triamcinolone acetonide, 0.1% twice daily for two weeks. Meanwhile, all his silver amalgam restorations were replaced with Glass Ionomer Cement (GIC). He was also advised to stop the habit of betel quid pouching and counselled for tobacco cessation. At two weeks, follow-up the patient was symptom-free with VAS score 0 [Table/Fig-3a,b] [1]. Medication was stopped and the patient was reassured and explained about the complications associated with this lesion The patient was asymptomatic at six months of follow-up [Table/Fig-4a,b] and is under regular monthly review.

DISCUSSION

Lymphangiectasia also called as acquired lymphangioma develops as a result of an abnormality to previously normal lymphatic drainage due to surgery, trauma, malignancy or radiotherapy [2]. The prevalence rate is 4% of all vascular tumours and has neither racial nor gender predilection [3]. Lymphangiomas are more common at birth or developed within 20 years of age, due to improper



[Table/Fig-3]: Review of the patient after two weeks topical application of triamcinolone acetonide. Follow-up images of (a) Tongue; and (b) Left buccal sulcus.



[Table/Fig-4]: Intraoral photograph at follow-up visit after six months of (a) tongue; and (b) left buccal sulcus.

connection of lymphatic channels to the main lymphatic drainage duct, while the acquired form often presents in adulthood as a sequelae of chronic lymphoedema, that leads to disruption as these cells do not anastomose efficiently with bigger lymphatic vessels, provoking areas of lymphatic blockage [4,5]. Approximately, half of all lesions are detected at birth and 90% develop by the age of two years [6].

The site of occurrence is often the head and neck region. Considering the oral cavity, most common site includes anterior two-thirds of the tongue, but infrequent occurrence has been reported in the buccal mucosa, lips, gingiva, palate and alveolar ridge of the mandible [4,6]. Superficial lesion represents papillary lesions with pebbly surface due to the occurrence of several translucent vesicles with same colour as that of adjacent mucosa or occasionally with a mild reddish hue. Interestingly, these give a tapioca pudding or frog eggs-like appearance [4,7]. The present case also presented in buccal mucosa and floor of mouth as a pigmented macule with a reticular pattern, unlike the usual exophytic growth, which has been reported in the literature to be relatively rare oral presentation [8,9]. The unilateral presentation of greyish radiating striae interspersed with hyperpigmentation mimicking lichenoid contact reaction was solely noted, which is the unique feature in this case; and to the best of authors' knowledge, first to be reported in the literature.

Mucosal pigmentation can be caused by long-term inflammatory mucosal disorders such as oral lichen planus, pemphigus, or pemphigoid [10]. The pathophysiology of postinflammatory pigmentation is unknown; however, it is more common in dark-skinned people [11]. Multiple brown-black pigmented patches are observed clinically proximal to reticular, erosive or vesicular lesions. Microscopically, melanin laden macrophages accumulate in the superficial connective tissue and there is excessive production of melanin by the melanocytes. Though, the clinical manifestations were consistent with the current instance, the histological findings did not support the diagnosis [10].

The oral melanotic macule is a tiny, well-circumscribed brown-to-black spot that most usually appears on the vermilion zone of the lower lip (33%), followed by the buccal mucosa, gingiva, and palate [6]. Histologically, it is marked by enhanced melanin synthesis with normal morphological characteristics. In the current case, both clinical and histological features varied, disqualifying the diagnosis.

In tobacco induced hyperpigmentation, a well-defined and predictable lesion develops at the place where the smokeless tobacco is retained in the oral cavity [12]. This type of pigmentation is present in 15% of tobacco chewers and caused by the irritational components in the tobacco, either because of stimulation of melanin generation or due to binding of melanin to the noxious components in the tobacco [12]. A similar clinical presentation was noted in the present patient as he had the habit of pouching betel quid in left buccal sulcus, but the absence of microscopic finding of increase melanin synthesis in the basal cell layer ruled out the diagnosis [12]. Mixed cell subepithelial infiltration, a deeper diffuse distribution in lamina propria, localised parakeratosis, focal disruption of the granular layer, and cytoid structures in the cornified and granular layers are histological markers of lichenoid lesions. This eliminated the preliminary diagnosis in the current case [13].

The pathogenesis of vessel proliferation in acquired form of lymphatic malformation occurs gradually in stages. Initially, the accumulation of interstitial fluid leads to swelling of the extracellular matrix, to which the endothelial cells lining the lymphatic capillaries become attached; following which the endothelial cells lining the lymph ducts become elongated at the capillary level, β-1-integrin, resulting in activation and phosphorylation of Vascular Endothelial Growth Factor (VEGF) receptor-3 [14,15]. This process results in the proliferation of endothelial cells in the lymph ducts, which is increased by oedema and mechanical pressure [14,15]. It is thought that, the inciting events in the index patient were the oedema caused by repeated trauma to the buccal mucosa and the mechanical pressure exerted by pouching the betel nut. A similar causative factor of trauma associated with trauma due to the mandibular space maintainer was reported previously in a case of lymphangiectasia, mimicking mucocele in lower labial mucosa [16].

Lymphatic vessels with significant dilatations are histopathologic characteristics of lymphangiectasia. The endothelial lining is thin, and the spaces are filled with proteinaceous fluid and lymphocytes. Secondary haemorrhage in lymphatic vessels is likely to occur. Lymphatic fluid, red blood cells, lymphocytes, macrophages and neutrophils are mostly found in the lymphatic spaces. Similar presentation of thin stratified squamous epithelium with vacuolar spaces and plenty of dilated endothelial lined lymphatic channels was noted, affirming the diagnosis [17].

The consequences of deep-seated lesions include obstruction of upper airway, extrusion of tongue, increased salivation, jaw deformity, pain, poor oral hygiene, difficulty in chewing and speaking [14]. The management depends upon their type, size, involvement of the structures and infiltration to the surrounding tissues [14]. Commonly, employed treatment modalities include laser therapy, cryotherapy, electrocautery, sclerotherapy, radiofrequency ablation and surgical excision [4]. Traditionally, treatment entails surgically removal without endangering crucial structures [18]. However, some adverse effects such as nerve injuries, recurrence, secondary infections, scar formation and incomplete resections because of involvement of adjacent vital structures [19].

Radiofrequency ablation used in a patient with oral lymphangioma showed no recurrence at one year follow-up. The present authors chose radiofrequency ablation because of the advantages of the technique to reach precise areas of tissue involvement, and being minimally invasive [8]. Surgical excision was done for an exophytic growth in a 30-year-old patient, which showed no recurrence at one year follow-up [9]. One of the two previously reported oral lymphangiectasia case associated with Crohn's disease, was treated successfully with short burst cryotherapy [20]. Radiation, laser and sclerotherapy have been associated with low success rates with potential side-effects [8]. As the index patient had only superficial lesion, the patient was symptomatically managed and explained the probability of adverse outcomes requiring close follow-ups.

CONCLUSION(S)

Acquired lymphangiectasia is rarely encountered in the oral cavity. Their timely recognition allows commencement of treatment with adequate follow-ups to prevent morbid states and recurrence. The case was exceptional due to the aetiology of trauma and the clinical appearance mirroring the lichenoid contact reaction. Consequently, while addressing such superficial mucosal lesions, acquired lymphangiectasia must be taken into account in the differential diagnosis. Fundamental knowledge about this condition is utmost importance, for its right diagnosis and proper therapeutic indication.

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