*Journal of Advances in Medicine and Medical Research*

***Volume 36, Issue 8, Page 84-92, 2024; Article no.JAMMR.120364***
***ISSN: 2456-8899, NLM ID: 101711724***
*(Past name: British Journal of Medicine and Medical Research, Past ISSN: 2231-0614, NLM ID: 101570965)*

# A Burning Issue on Oral Mucosal Diseases: Case Series with Review

**Durba Roychowdhury [a++*], Rudra Prasad Chatterjee [a,b#], Sk. Abdul Mahmud [a†], Sudeshna Bagchi [a‡] and Arunit Chatterjee [a‡]**

*[a] Department of Oral and Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Kolkata, West Bengal, India.*
*[b] Department of Oral and Dental Sciences, JIS University, Kolkata, India.*

***Authors' contributions***

*This work was carried out in collaboration among all authors. Author DR designed the study, performed the statistical analysis, wrote the protocol, and wrote the first draft of the manuscript. Authors RPC, SK.AM and SB managed the analyses of the study. Author AC managed the literature searches. All authors read and approved the final manuscript.*

***Article Information***

DOI: https://doi.org/10.9734/jammr/2024/v36i85528

**Open Peer Review History:**
This journal follows the Advanced Open Peer Review policy. Identity of the Reviewers, Editor(s) and additional Reviewers, peer review comments, different versions of the manuscript, comments of the editors, etc are available here: https://www.sdiarticle5.com/review-history/120364

***Case Report***

***Received: 25/05/2024***
***Accepted: 23/07/2024***
***Published: 27/07/2024***

## ABSTRACT

Oral Lichenoid Reaction (OLR) is a chronic inflammatory lesion of the oral mucosa that occurs as an allergic response to certain dental materials, medications and systemic diseases. The frequency of OLR in the general population has been documented to be very less. The clinical and histological features of OLR closely resemble those of Oral Lichen Planus (OLP), making it

*[++] (BDS), Post Graduate Trainee;*
*[#] MDS), PhD Scholar & Professor;*
*[†] Professor;*
*[‡] (MDS), Assistant Professor;*
**Corresponding author: E-mail: durbaroychowdhury513@gmail.com;*

***Cite as:*** *Roychowdhury, Durba, Rudra Prasad Chatterjee, Sk. Abdul Mahmud, Sudeshna Bagchi, and Arunit Chatterjee. 2024. "A Burning Issue on Oral Mucosal Diseases: Case Series With Review". Journal of Advances in Medicine and Medical Research 36 (8):84-92. https://doi.org/10.9734/jammr/2024/v36i85528.*

challenging to distinguish between the two clinically. OLRs might have a higher malignant potential than OLP. The diagnosis and treatment of OLR is very crucial as misdiagnosis may result in detrimental effects on the biophysical health of the patient. Here, we present case series of two patients who presented with amalgam restorations and burning sensation on the buccal mucosa while consumption of hot and spicy food. An immunohistopathological evaluation confirmed the diagnosis as Oral Lichenoid Reaction. Elimination of causative factors remain the mainstay of treatment which markedly reduces the sufferings of the patient with a commendable result.

*Keywords: Lichenoid reaction; DIF; burning; mucous membrane; amalgam.*

## 1. INTRODUCTION

The oral mucosa provides effective barrier which is often in constant contact with various deleterious substances, acidic or alkaline compounds, spicy or non-spicy food, allergens like chemical products used in toothpastes, mouthwashes, oral flavoring agents, preservatives and dental materials [1]. Oral lichenoid reaction (OLR) is characterized as a persistent inflammatory condition affecting the oral mucosa, triggered by allergic reactions to dental products, specific medications, individuals with graft-vs-host disease (GVHD) and systemic conditions [2]. Histologically, OLRs mimic oral lichen planus (OLP), thus, clinical interpretation is mandatory [3]. The occurrence of OLR is a common phenomenon, manifesting with a prevalence of 2.4% among the general populace [2]. These lesions most commonly occur in middle-aged adults with a slight female predilection [4]. Areas commonly involved are the buccal mucosa, lateral border of the tongue and labial mucosa that are in direct contact with metal restorations or other allergens. It is generally limited in size and unilateral in distribution. On the contrary, OLP lesions are frequently found bilaterally in the oral mucosa which serve as the distinguishing feature. Clinically, OLP shows a wide range of variations, from white interlacing striations to ulcerations even blister formation, and are asymptomatic in nature. Patients may complain of burning sensation along with intolerance to spicy meals [2]. Based on an admixture of clinical diagnosis, histopathological evaluation and immunofluorescence test, a final diagnosis was made and patients were successfully treated.

## 2. CASE PRESENTATION

### 2.1 Case 1

A 61-year-old female patient from a semiurban area reported to the Department of Oral and Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Kolkata with the chief complaint of mild burning sensation over left buccal mucosa, lower labial mucosa and tongue since last 1-2 years, which was small initially but progressively increased over time to the present size (measuring about 1.5x1.2 cm) being associated with burning sensation on taking hot and spicy food. At the time of presentation, the patient had a class I amalgam restoration on 38 that was performed approximately 6 years ago which was now considered as poor or defective. However, the affected site was free from traumatic occlusion or from sharp cuspal edges of tooth or dentures. The medical history revealed that she was hypertensive and under medication. Intraorally, the presence of inflamed and erythematous attached gingiva and interdental papilla with respect to 31,32,33 and 41,42,43 tooth region [according to Federation Dentaire Internationale (FDI) notation] was observed. Gingival recession was noted wrt 31,32 and poor oral hygiene is present. There was also presence of small erythematous ulcerated lesion over gingivobuccal sulcus and buccal mucosa with respect to 36 [FDI notation]. The patient also had a fissured depapillated tongue [Fig. 1]. These led to a provisional diagnosis of oral mucositis.

Thereafter, the patient was advised for oral prophylaxis along with empirical antibiotic therapy and antibacterial mouthwash. Most of the lesions started to regress after 1 month of therapy but the lesion adjacent to 36 was still persistent and the incisional biopsy was planned from that representative area.

The patient was then advised to undergo routine hematological and biochemical investigations which were within normal limits. Incisional biopsy was performed under local anesthesia and submitted in buffered formalin for routine histopathological examination. Sections stained with Haematoxylin & Eosin revealed the

presence of hyperplasia of surface epithelium with basal cell degeneration. Spongiosis can be encountered due to presence of intercellular edema. Subepithelial stroma showed numerous diffuse chronic inflammatory cells infiltrate extending from juxta epithelial area to deep into the connective tissue accompanied by plasma cells and histiocytes. No cellular atypia was evident [Fig. 2]. The light microscopic features were suggestive of lichenoid reaction.

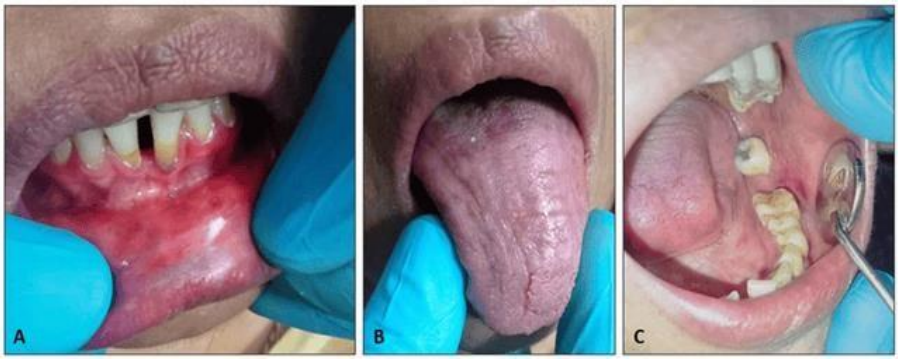


**Fig. 1. Intraoral photograph of the patient showing (A) presence of inflamed and erythematous attached gingiva and interdental papilla with respect to 31,32,33 and 41,42,43 tooth region; (B) Fissured depapillated tongue; (C) Small erythematous area over gingivobuccal sulcus and buccal mucosa wrt edentulous space of 37**

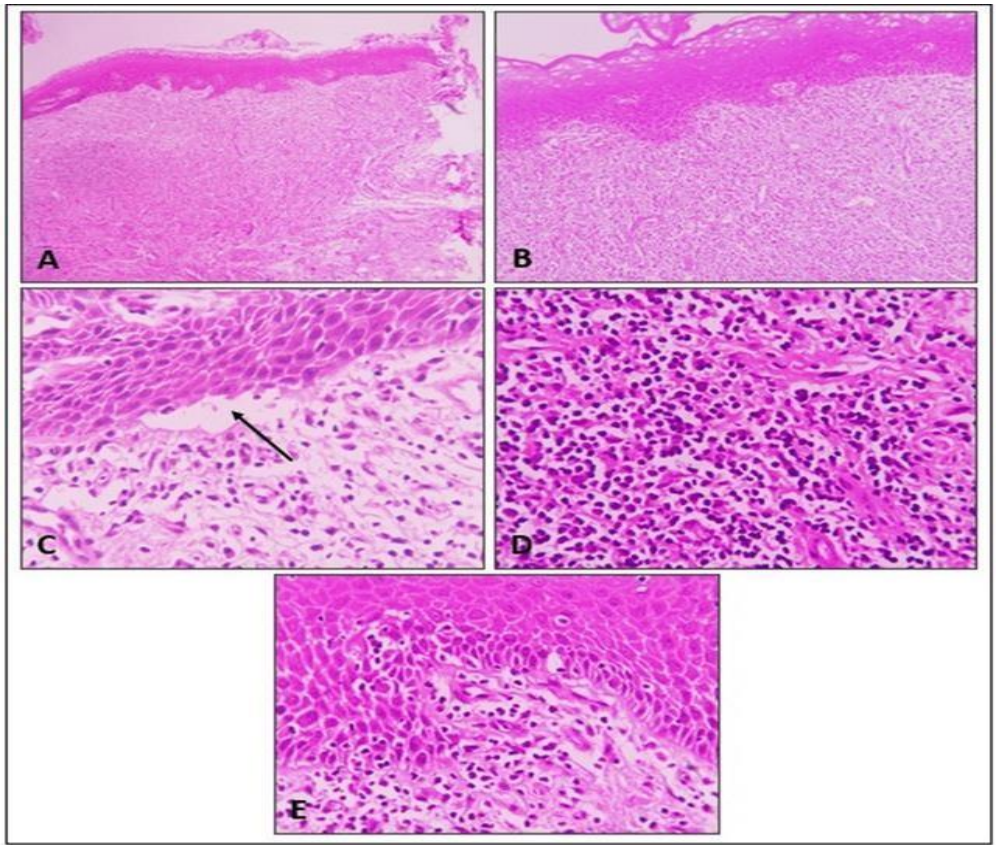


**Fig. 2. (A) Photomicrograph showing hyperplastic stratified squamous surface epithelium with underlying connective tissue stroma (H and E; 4X); (B) Spongiosis and diffuse chronic inflammatory cells infiltrate deeper into the lamina propia (10X); (C, D, E) Max Joseph spaces (arrow) and basal cell degeneration; numerous chronic inflammatory cells infiltrate chiefly characterized by lymphocytes, histiocytes along with plasma cells (40X)**

To confirm the diagnosis, direct immunofluorescence (DIF) was advised. A perilesional tissue was taken and sent for the DIF testing directly in Michel's medium, which revealed no appreciable staining deposits for IgG, IgA, IgM and C3 [Fig. 3].

The overall clinical, histopathological features and immunofluorescence study were consistent with lichenoid inflammation.

The treatment included elimination of the amalgam filling in the lower back tooth region by Glass Ionomer restoration and application of topical clobetasol (0.05% w/w) and miconazole (2% w/w) on the affected areas along with benzydamine (0.15% w/v) mouthwash and followed for 2 months. The patient responded well to the above-mentioned treatment and no further exacerbations were noticed [Fig. 4].

## 2.2 Case 2

A 57-year-old male patient reported to the Department of Oral and Maxillofacial Pathology with a chief complaint of burning sensation in his cheeks while consumption of hot and spicy food for the last 1 year. The patient visited to dental surgeon for similar complains 6 months ago and applied steroid ointment as per prescription. Remission of the lesion was observed after 4 months of treatment but recurrence was noted over a period of 2 months after termination of medication. Medical history revealed that the patient was hypertensive under medication. On intraoral examination, we noted the presence of white striations with central erythema on bilateral buccal mucosa. Amalgam restoration in the left mandibular second molar and right mandibular third molar was done approximately 3-4 years ago. The rest of the oral mucosa appeared to be normal [Fig. 5].

A provisional diagnosis of oral lichen planus was made based on the clinical findings. The patient's hemogram was within normal limits. Incisional biopsy was performed under local anesthesia from the representative site and finally sent for routine light microscopic histopathological evaluation to confirm a diagnosis.

Sections stained with H&E revealed the presence of stratified squamous surface epithelium with irregular rete ridges, focal acantholysis and occasional basal cell degeneration. Underlying stroma revealed diffuse chronic inflammatory cell infiltrate extending deeper into the connective tissue layer with engorged blood vessels and large number of eosinophils. No signs of malignancy could be detected [Fig. 6].

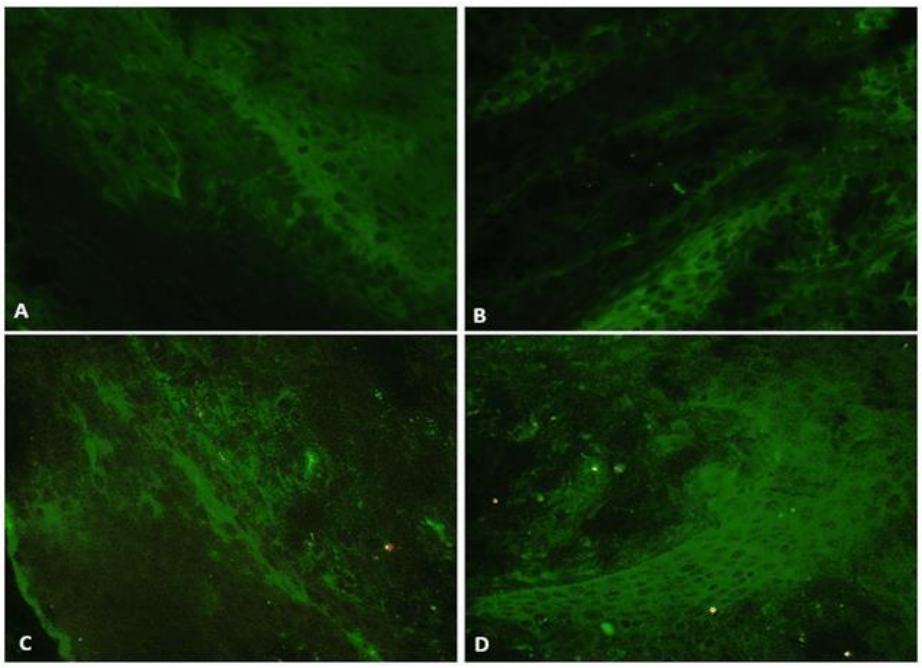


**Fig. 3. Direct Immunofluorescence (DIF) showing a negative staining deposits for (A) IgG (B) IgA (C) IgM and (D) C3**

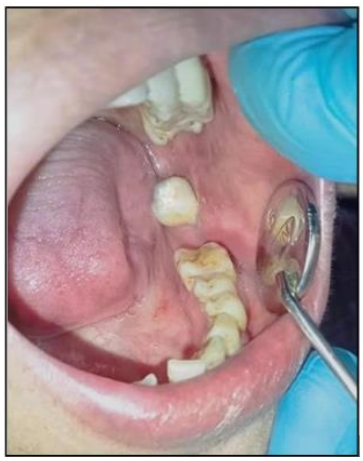

**Fig. 4. Post-treatment after 2 months**

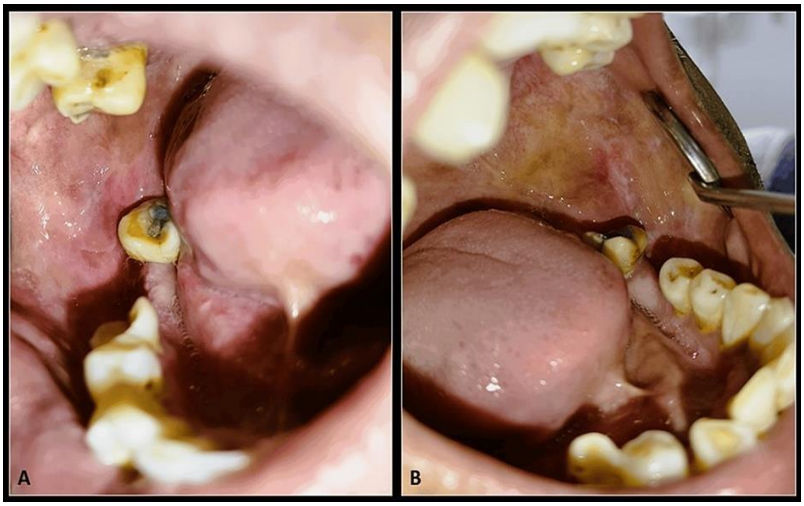


**Fig. 5. (A, B): Intraoral examination showing presence of white striations with central erythema on bilateral buccal mucosa adjacent to teeth wrt 37 (left mandibular second molar) and 48 (right mandibular third molar) filled with amalgam**

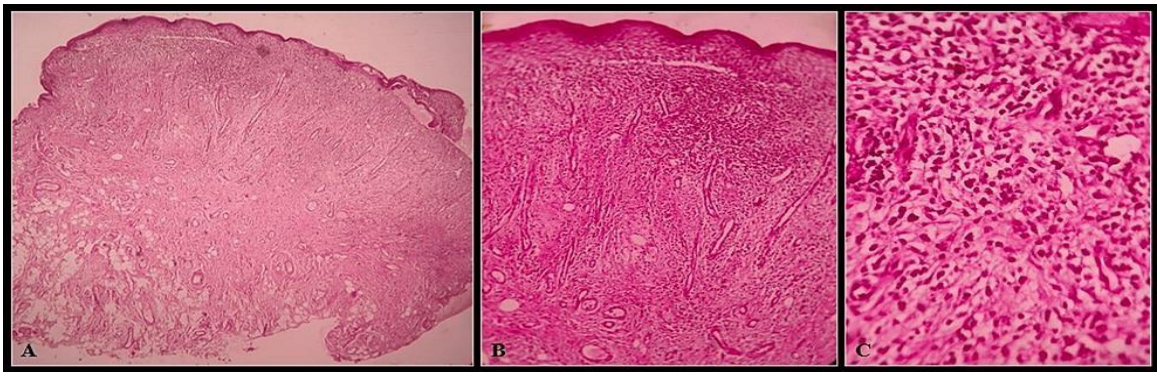


**Fig. 6. (A TO C) Photomicrograph showing hematoxylin and eosin-stained sections revealing stratified squamous epithelium with irregular rete ridges (4X); Focal acanthosis, occasional basal cell degeneration and chronic inflammatory infiltrate deeper into connective tissue layer (10X); Inflammatory infiltrate along with engorged blood vessels and large no of eosinophils (40X)**

Thus, the diagnosis of oral lichenoid reaction was made based on histopathological evaluation. The patient was then advised to replace the amalgam restorations with a tooth-colored restorative material and composite resin restoration was done. His symptoms regressed and complete healing was noted after 2.5 months. There has been no evidence of recurrence or discomfort over a period of 1 year. The comparative analysis of both the patients given in Table 1.

## 3. DISCUSSION

Pinkus first introduced the term "Oral Lichenoid Tissue Reaction" in 1973 to describe the histological pattern indicating damage to keratinocytes along with the infiltration of inflammatory cells in the connective tissue that may also extend into the epithelium [1]. Oral Lichenoid Tissue Reaction has also been termed as Oral Lichenoid Lesions (OLL), Oral Lichenoid Reaction (OLR), lichenoid contact stomatitis, or lichen-planus-like lesions due to the clinical and histological similarities between OLR and Oral Lichen Planus [2]. OLRs can manifest either as a distinct pathological condition or as an exacerbation of pre-existing oral lichen planus. According to Van der Waal (2009) OLRs can be categorized into four types: Amalgam restoration topographically associated lesions, Drug-associated lichenoid lesions, Lichenoid lesions in individuals having Chronic Graft Versus Host Disease and lesions that have a lichen planus like aspect but that lack one or more characteristic clinical aspects [1].

Lichenoid reactions have been attributed to several drugs such as Beta Blockers, Nonsteroidal Anti-Inflammatory Drugs (NSAIDs), Dapsone, Oral Hypoglycemics, Penicillamine, Sulfonylureas, and Anti-Psychotic like Phenothiazines, Vasodilators. They have also been associated with dental materials including amalgam, dental acrylics etc. [4,5]. Association of systemic diseases such as Chronic Hepatitis C and patients vaccinated against Hepatitis B have been observed in numerous cases [6]. In our case, one of the patients had the amalgam restoration on 38 (class I) while the other had on 38 and 48 respectively.

**Table 1. Comparative analysis of both cases with following criteria**

| Features | Case 1 | Case 2 |
|---|---|---|
| Age & sex | 61-year-old female patient | 57-year-old male patient |
| Type of reaction | Delayed | Delayed |
| Allergen location | Amalgam restoration on 38 | Amalgam restoration on 38 and 48 respectively. |
| Duration of contact | 6 years | 3-4 years |
| Burning sensation | Present | Present |
| Erythema | Present | Present |
| White striations | Absent | Present |
| Site | Buccal mucosa vicinity to amalgam restoration | Buccal mucosa on both sides vicinity to amalgam restoration |
| Histological features | Diffuse inflammatory infiltrate extending deeper into the lamina propria; basal cell degeneration; numerous chronic inflammatory cellsinfiltrate chiefly characterized by lymphocytes, histiocytes along with plasma cells | Diffuse inflammatory infiltrate extendingdeeper into the lamina propria. Occasional basal cell degeneration and chronic inflammatory infiltrate along with engorged blood vessels and large no ofeosinophils |
| DIF | No appreciable staining deposits for IgG, IgA, IgM and C3 | Not performed |
| Treatment | Amalgam restoration was replacedby a tooth-coloured restorative material like composite resin. | Amalgam restoration was replaced by atooth-coloured restorative material like composite resin. |
| Follow-up | Started experiencing relief after 3months of therapy | Started experiencing relief around 2.5 months and complete healing was noticed in subsequent follow-up |

The pathogenesis of OLRs is still largely unknown. It is postulated that various pathways of antigen presentation could serve as a fundamental determinant. At the time of restoration, OLRs are barely encountered due to insolubility of amalgam into the saliva and its washing mechanism [7]. The response initiates when haptens (incomplete antigens, combine with proteins/counterparts to create full antigens) interact with the oral mucosa. Following the first encounter, an initial local immune and inflammatory response takes place and the antigen internalized by macrophages and monocytes is subsequently displayed to T cells leading to their sensitization and activation of CD4+ T cells. Upon subsequent exposure to the identical allergen, these cells secrete cytokines and chemokines that have the potential to incite an immune reaction against epithelial antigens, thereby instigating the formation of OLR and this seems to manifest as a T-cell mediated delayed hypersensitivity reaction (Type IV) upon contact with either the mercury or another constituent of amalgam. This response could be delayed for a minimum 48 hours and the manifestation of symptoms may vary based on the severity of the reactions which can manifest as either be acute or chronic [8-11]. Previous studies demonstrated that the expression of nuclear factor kβ - dependent cytokines in serum, oral keratinocytes and tissue-infiltrated mononuclear cells including TNF-α, IL-1 and IL-6 was increased in individuals with OLR [5].

The prevalence of LR has been reported to be approximately 2.4% in the general population and middle-aged individuals are commonly affected [2]. In our case series, our patients also belonged to the mentioned age group.

Clinical presentations may vary depending on the type of reaction, allergen location and duration of contact. The asymmetry of the lesion is notable; however, it can be present bilaterally if there are amalgam restorations on both sides. The buccal mucosa is the most frequently affected site followed by border of tongue [5]. Acute lesions may present with symptoms such as burning sensation and redness. Vesicles are not commonly observed, but if present, they tend to rupture shortly after forming, leading to the presence of erythematous areas. Chronic lesions typically manifest as regions of erythema, oedema, desquamation and occasionally ulceration [11]. Both the patients had a lesion over the buccal mucosa which was consistent with the clinical presentation documented in the existing literature. In the 2nd case, the patient had white striations with erythema in certain areas of buccal mucosa which was clinically corroborative with the features of OLP.

Histopathologically, there is the presence of hyperkeratosis of surface epithelium along with spongiosis, liquefactive degeneration of the basal cell layer and diffuse inflammatory infiltrate extending deeper into the lamina propria unlike OLP where chronic inflammatory cells are restricted to the juxta-epithelial connective tissue in a band like fashion. This infiltrate comprises of plasma cells and eosinophils in addition to lymphocytes and increased numbers of colloid or civatte bodies in case of OLR. Perivascular chronic inflammatory cell infiltrate may be seen in drug related lichenoid lesions [12]. Our cases showed similar histologic presentations. Since, in both the cases, there is presence of chronic inflammatory cell infiltrate extending deeper into the connective tissue layer and large number of eosinophils, the possibility of OLP is excluded.

Clinically and immunohistopathologically, differential diagnosis of OLRs include: Vesiculo-Bullous Diseases such as Pemphigus Vulgaris, Leukoplakia, Lupus erythematosus (LE) etc. [11]. Histopathological evaluation of bullous diseases reveals intraepithelial, subepithelial and suprabasilar split. We can also exclude Leukoplakia from OLR as Leukoplakia clinically appears as greyish-white with cracked mud appearance and shows dysplastic epithelium [12]. All these features were missing in our cases.

Direct Immunofluorescence (DIF) was used to examine the fluorescence patterns in oral lichenoid reactions and to compare the degree of intensity of their fluorescence [13]. It was observed that the immunofluorescence pattern of OLP is more ragged and fibrillary whereas it is more uniform and less intense in OLR. However, fibrinogen deposition at the basement membrane zone was absent in 29.2% of OLR [3]. No appreciable staining was also noted in our case.

In case of Lupus, DIF of lesional tissue typically reveals the presence of one or more immunoreactants (usually IgM, IgG, or C3) forming a shaggy or granular band at the basement membrane zone [14] which was negative in our cases.

The confirmation of pemphigus vulgaris diagnosis necessitates the direct immunofluorescence evaluation of the

perilesional tissue, demonstrating the presence of antibodies (usually IgG or IgM) and complement components (usually C3) in the intercellular spaces between the epithelial cells, characterized by a pattern resembling a fish net or chicken wire which is not present in our case series [15].

In the present case series, we pinned our confirmatory diagnosis as Oral lichenoid reaction based on clinical findings, histopathological features and diagnosis of exclusion.

Remission of OLR involves the substitution of causative restorations with non-allergic material. Replacing amalgam restorations has resulted in significant enhancements in 93% of hypersensitivity lesions associated with amalgam contact [16]. In both the cases, the amalgam restoration was replaced by a tooth-coloured restorative material like composite resin. The patient started experiencing relief around 2.5-3 months and complete healing was seen in subsequent follow-up. There has been some controversy about the malignant potential of lichenoid reactions (2.43%) which is usually thought to be extremely rare [1,17]. Iocca et al. (2020) stated that the true potential of the malignant transformation of OLLs is high (3.8%) [18]. However, the patients should be regularly monitored until the complete remission of the lesion is noted [1].

## 4. CONCLUSION

Dental amalgam continues to be the most preferred and affordable restorative material in the field of restorative dentistry before a decade despite the availability of new synthetic non-metallic alternatives, predominantly due to its superior strength as well as minimal technique sensitivity. But it might result in oral lichenoid reaction in susceptible patients. When the lesion appears in the close proximity to amalgam restoration, replacement of such restoration can be eliminated by substitution with alternative tooth colored restorative material like Glass ionomer or composite resins are recommended.

Although the clinicopathological characteristics of OLP are similar, OLR and OLP must be distinguished from one another due to differences in etiology, diagnosis and prognosis, failure to do so may result in detrimental effects to the patient. OLRs exhibit a high susceptibility to undergo malignant transformation. So, early diagnosis and periodic follow-up represent crucial measures in impeding the progression of the condition.

## DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of manuscripts.

## CONSENT

As per international standards or university standards, written informed consent has been obtained from the patient (or other approved parties) and preserved by the author for publication of this case report and accompanying images.

## ETHICAL APPROVAL

It is not applicable.

## ACKNOWLEDGEMENTS

We are thankful to all the faculty members, postgraduate students, and other staff of the Department of Oral and Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, for their immense support and contributions from time to time. Our sincere gratitude to Prof. (Dr.) R.R. Paul, Prof (Dr.) Mousumi Pal, Dr. Neha Shah for their immense help and relentless support. Words are inadequate to express gratitude to them. There is no conflict of interest related to this study.

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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*The peer review history for this paper can be accessed here:*
*https://www.sdiarticle5.com/review-history/120364*