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CASE REPORT

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Oral lichen nitidus case report

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Key Clinical Message

This case report aims to increase awareness that lichen nitidus may affect the mouth and therefore supports multidisciplinary management, particularly between dermatologists and dental professionals.

KEYWORDS

case report, dermatology, lichen nitidus, oral, oral medicine, oral pathology

1 | INTRODUCTION

Lichen nitidus (LN) is an uncommon inflammatory skin condition of unknown aetiology. LN can involve the nails.[1] Oral involvement is rare with only one oral biopsy-proven case of oral LN in the literature,[2] which presented as white papules on the dorsal tongue. Four other papers have documented cases of oral lesions presenting in patients with biopsy-proven cutaneous LN.[1,3-5] Like oral lichen planus (OLP), LN is a condition driven by a lichenoid inflammatory process. Cutaneous LN typically presents in childhood or adolescence with asymptomatic eruptions of small skin-colored/yellow papules, particularly on the flexor aspects of the limbs, abdomen, and genitals. LN has distinct clinical and histopathological features, but the pathophysiology of LN is poorly understood.

We present a case, that was referred to our Oral Medicine and Oral & Maxillofacial Surgery departments, of biopsy-proven oral LN prsenting with red, erythematous papules on the palatal mucosal.

2 | CASE HISTORY AND EXAMINATION

A 64-year-old woman presented with 5-year history of raised areas affecting the roof of her mouth. She reported a single episode of severe discomfort from the site, otherwise her mouth was largely asymptomatic. Mild irritations were relieved by changing to a sodium-lauryl sulfate (SLS) free toothpaste. There was no history of any trauma or injury to the palatal mucosa. Topical and systemic antifungal treatment made no difference to the lesions. Medically, she reported a possible previous diagnosis of vulval lichen sclerosis. The patient was a lifelong nonsmoker, drank 6 units of alcohol per week, and had no history of recreational drug use. The patient denied having any skin lesions and a comprehensive skin examination was not undertaken. Intra-oral examination revealed a depapillated dorsum of tongue and multiple discrete papules of 2 mm in greatest dimension affecting the hard and soft palate (Figures 1 and 2). Some of these papules were erythematous in nature

and extended to involve the uvulae and pillars of fauces. Otherwise, all other soft tissues were healthy.

3 | INVESTIGATIONS AND DIAGNOSIS

Preliminary investigations were undertaken at a local Oral and Maxillofacial Surgery department. As reported by a general pathologist, histopathological findings from the initial biopsy taken from the hard palate were non-specific with "relatively heavy ductal/periductal chronic inflammation of the minor salivary gland ducts associated with slight lobular hyperplasia and slight keratosis."

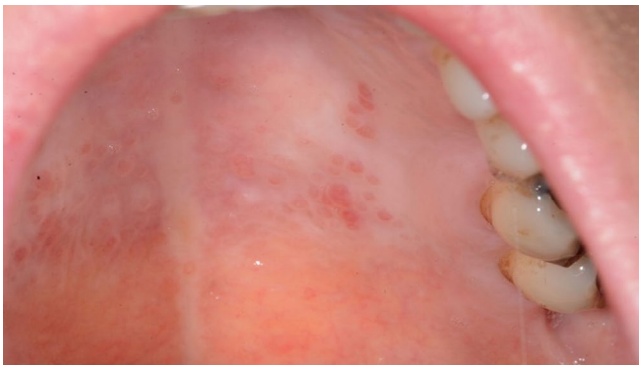

FIGURE 1 Clinical picture showing multiple well-defined erythematous circular papules affecting the palatal mucosa

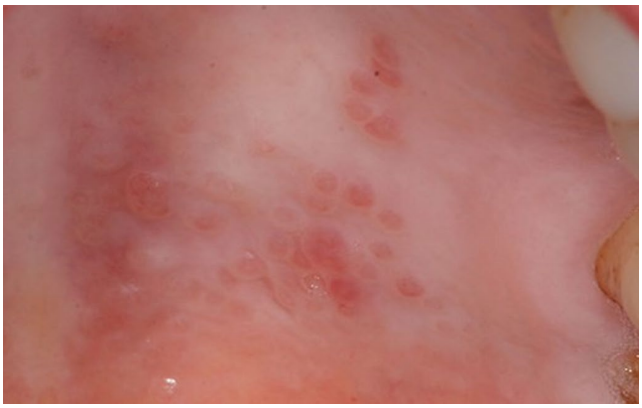

FIGURE 2 Clinical picture showing multiple well-defined erythematous circular papules affecting the palatal mucosa

TABLE 1 Differential diagnoses that were considered but deemed very unlikely

Differential diagnosis
Papillary hyperplasia of the palate
Candidiasis
Stomatitis nicotina

Examination of further incisional biopsies of two separate sites of the lesions on the palatal mucosa were reported as patchy chronic inflammation with lichenoid features and prominent minor salivary gland tissue.

Routine bloods were taken including full blood count, hematinics, thyroid function, and HbA1c all of which were normal.

A referral to Oral Medicine was made, and the patient was seen in the Oral Medicine department. Given that the appearance of the papules was very unusual, a clinical diagnosis could not be made. The differential diagnoses listed in Table 1 were considered but deemed very unlikely (see Table 1). A swab was taken from the affected area, which did not yield any significant growth of Candida or pathogenic bacteria. Furthermore, the appearance of the papules was not typical of candidiasis and the patient had no local or systemic factors predisposing to candidiasis. The patient was a lifelong nonsmoker, therefore making stomatitis nicotina an extremely unlikely diagnosis. Papillary hyperplasia of the palate was considered but this patient was not a denture wearer, nor was she taking any medications that have been implicated in gingival hyperplasia.

Therefore, the histopathology slides and additional levels of the more recent palatal biopsies were reviewed by a consultant oral pathologist who recognized that while there was a dense band of inflammatory cell infiltrate adjacent to the epithelium, the histopathological features were not those of OLP and suspected LN. As LN is much more commonly seen on the skin, a second opinion was sought from a consultant dermatopathologist who agreed with the diagnosis of LN. The histopathological appearances were characteristic of LN with small raised nodules, lichenoid inflammation, and a "claw clutching ball" configuration (see Figures 3-5). While the inflammatory changes are similar in OLP, the architectural features observed are not evident in OLP.

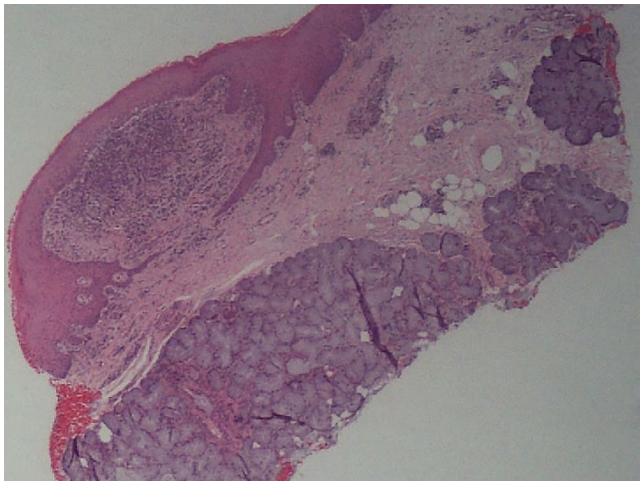

FIGURE 3 (H& E × 20): A small well-defined raised surface lesion is seen

4 | MANAGEMENT AND FOLLOW-UP

No active treatment was prescribed as the lesions were largely asymptomatic. The patient was reviewed in Oral Medicine at 6 months, and the lesions were unchanged. The patient was given the diagnosis, reassured, and provided with an open appointment.

5 | DISCUSSION

Oral lichen nitidus is uncommon: a literature search yielded only five reported cases in the last 91 years that suggested LN in the differential diagnosis.[1-5] Only one of these supported this diagnosis with a biopsy of the oral lesions.[2] The other four cases reported skin biopsy-proven LN in patients who also had mucosal lesions suspicious of LN (e.g. yellow papules on gums[4]). LN and OLP have been reported to occur concurrently,[6] and in two of the above cases, OLP was also suggested in the differential diagnosis.[3,4]

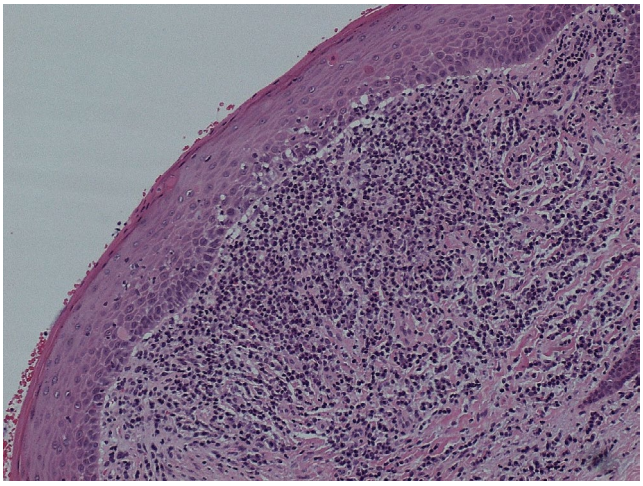

FIGURE 4 (H&E × 100): Shows marked lichenoid inflammation

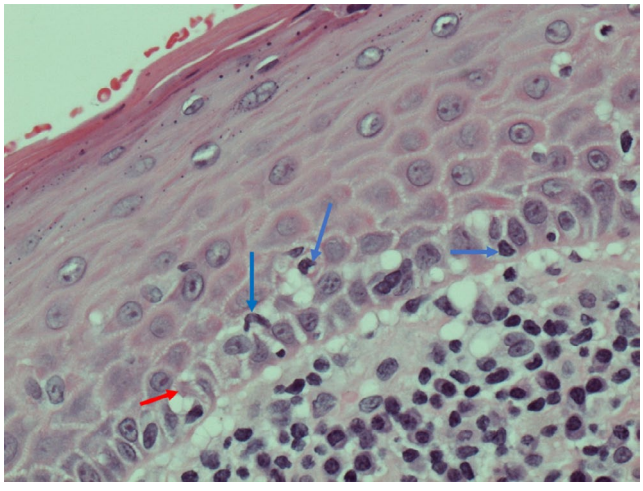

FIGURE 5 (H&E × 400): High-power view showing lymphocytic exocytosis (blue arrows) and Civatte bodies (red arrow)

The present case presented clinically with intra-oral red erythematous papules and histologically the epithelium over the central surface of the nodules was mildly atrophic with slightly less keratinization compared with the adjacent mucosa. In contrast, the previous case report of biopsy-proven oral LN presented clinically with white papules.[2] It is unclear why there is variation, but it is noted that cutaneous lesions can be variable in appearance. Cutaneous LN lesions are predominately skin-colored but in some cases can present red/brown in color[7] or even hemorrhagic[8] in appearance. Therefore, the red erythematous papules in this case could parallel this redder cutaneous variant.

The patient denied having any skin lesions. Therefore, the decision was made not to seek a dermatology opinion, as it was felt this would be unlikely to change their management. This raises the question as to whether all patients with rare oral mucosal lesions related to dermatological pathology should be examined by a dermatologist.

Historically, LN was believed to be a variant of lichen planus (LP) and some have argued that LN may be a preceding condition to LP. However, investigations have revealed differences between the immune-mediated responses in LP and LN, and LN is now considered by most to be a distinct condition.[9] For example, while the inflammation in both LP and LN is predominately driven by T lymphocytes, the proportion of these that are HECA-452+ is greater in LP compared with LN.[9] The exact etiology of LN has been a subject of debate. Cutaneous LN has been reported to occur in patients with alterations in their immune status such as in patients with advanced HIV disease[10] and those receiving systemic therapy with interferon-α and ribavirin for hepatitis C infection.[11] More recently, various inflammatory eruptions including LN[12] and LP[13] have been reported in patients receiving immune checkpoint inhibitor immunotherapy for advanced cancers.

These observations support the fact that LN likely develops in situations of immunological change. The case we describe, however, had not received any immunotherapy and did not suffer from any conditions linked to alterations in immune status. LN has been reported to occur concurrently with other conditions such as LP,[14] Crohn's disease,[15] and psoriasis vulgaris.[14] This patient did have a possible diagnosis of lichen sclerosis but no other significant findings so far as their medical history was concerned. Furthermore, the histological appearance of the intra-oral papules in this case was not those of lichen sclerosis.

It is possible that oral LN may present more commonly than is reported. In this case, the first and subsequent biopsies

were initially described as "chronic inflammatory changes" prior to the definitive diagnosis being made. This may relate to the lack of clinical suspicion of the condition in the mouth. It also highlights the need to examine several histological levels to identify the small focal lesions and the importance of clinicopathological correlation. LN has characteristic histopathological features: each papule is well-defined and can involve up to five connective tissue papillae. "Claw-like" extensions of rete ridges are present at the boundary of the papule. The inflammatory cellular infiltrate typically consists of lymphocytes, epithelioid cells, and histiocytes. Exocytosis of lymphocytes with basal cell degeneration is a feature, and Civatte bodies can be present in the basal layer.[16] The oral lesions in this case display these distinctive histological features.

Another contributing factor in the low rate of oral diagnosis may be the clinical presentation of oral LN itself. While the dermatosis seen in skin LN has clear documented clinical features, the clinical appearance of oral LN is poorly characterized. While it has been suggested that oral LN may appear similar to OLP[3] this case has an appearance distinct from any variant of OLP in particular, there was no clinical evidence of hyperkeratosis.

Since the cutaneous lesions of LN are usually asymptomatic, treatment is usually not necessary. In certain cases where patients experience a pruritus, therapies such as topical corticosteroids or light treatments (sunlight, narrowband UVB phototherapy and photochemotherapy (PUVA)) can be used. Systemic therapies are rarely required.

6 | CONCLUSION

Our findings in this case point to a diagnosis of oral LN. However, further cases need to be reported to confirm the existence of this variant. The unknown etiology of LN makes it difficult to say why this patient developed LN in the mouth. LN affecting the oral mucosa may be underreported and underdiagnosed. This could be a result of its often asymptomatic nature or a lack of knowledge of the existence of the condition among dental and other healthcare professionals. Increased awareness may lead to further cases being reported, confirming that this condition may present in the mouth. This may also foster a multidisciplinary approach in cases where oral and skin lesions co-exist, particularly if these are symptomatic and require active management.

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CONFLICT OF INTEREST

No potential conflict of interest is reported by the authors.

AUTHOR CONTRIBUTION

M L Dobson: conducted literature search on the topic and drafted and revised the paper. A Brown: conducted literature search on the topic and revised the paper. E D Theaker: supervised work and revised the paper. S J White: supervised work, led histopathology interpretation of case, linked this to the literature, and revised the paper.

DATA AVAILABILITY STATEMENT

The data for this case report were taken from the case clinical records and anonymized. Written patient consent was gained for this case report.

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