

CASE REPORT

Lichen Planus of Larynx Manifesting as Airway Compromise: A Rare Presentation

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ABSTRACT

Introduction: Lichen planus is an inflammatory pathology which most commonly involves the mucocutaneous junction of oral cavity. It has been reported in many other sites of the body extending from eye to the urogenital system. Diagnosis is achieved combinely with clinical picture and histopathological findings. The treatment mainly involves systemic therapy with corticosteroids and regular periodic follow-up. It is rare but malignant transformation has been suggested at some sites.

Case report: However, it is extremely rare in larynx and this case report seems to be the first case presenting with a combination of airway compromise due to laryngeal involvement with presence of cutaneous and oral lesions.

Conclusion: The above case report is presented in context of developing insights about etiopathogenesis, clinical features and management of laryngeal lichen planus.

Keywords: Clinical features, Lichen planus, Management, Oral cavity.

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INTRODUCTION

Lichen planus (LP) is a chronic inflammatory condition commonly affecting the oral mucous membranes. Its reported incidence is about 0.1 to 4% of the population; however topographical and racial variations exists.¹

Several other mucosal sites like oropharynx, esophagus, vulvovaginal region in females and the penis in males have been reported to be affected by lichen planus. Very rarely, it has been reported to involve larynx.²

Its clinical presentation may be variable, including reticular, papular, plaque-like, atrophic, and ulcerative lesions. Cutaneous involvement is seen on the flexor surface of the wrists, lower back, ankles and scalp. It is usually seen in middle-aged people, with a slight preponderance to females.¹

In this report, we describe a rare case of lichen planus affecting the oropharyngeal cavity and laryngeal mucosa with genital involvement. This seems to be the first case report of such association to be reported from India.

CASE REPORT

We report a case of a 45-year-old lady from Bhutan who presented to us with history of recurrent oral ulceration of 10 months duration. She also complained of breathlessness and noisy breathing associated with hoarseness of voice for 4 months. She was initially evaluated in Calcutta for the above complaints, where she was tracheostomised and thereafter referred to our center for further evaluation. She gives no history of dysphagia, odynophagia, hemoptysis, fever, weight loss, ear or nasal complaints.

General examination revealed that her vitals were stable; however mild inspiratory stridor was present. Oropharyngeal examination showed scarring of soft palate and tip of dorsum of tongue. Flexible laryngoscopy along with computerized tomography showed a web in the anterior 2/3rd of the glottis compromising the airway (Figs 1 and 2). Routine blood examination was normal. Her immunological workup including CANCA, PANCA and CRP was within normal limits.

She underwent microlaryngeal surgery and web excision using cold steel with a keel placement. Biopsy of the web showed mucosa, epithelium showing acanthosis, focal basal cell vacuolation and exocytosis. Scant subepithelium showing band like lymphocytic infiltrate and telangiectasia without any evidence of malignancy (Figs 3A and B). These features were consistent with lichen planus.

On further detail evaluation by dermatology, she was also found to have genital lesions consistent with lichen planus. She was prescribed a course of oral prednisolone (tapered dose) along with topical triamcinolone for a period of 6 months.

The procedure had to be repeated a second time after 6 months because of recurrence of the web. She underwent

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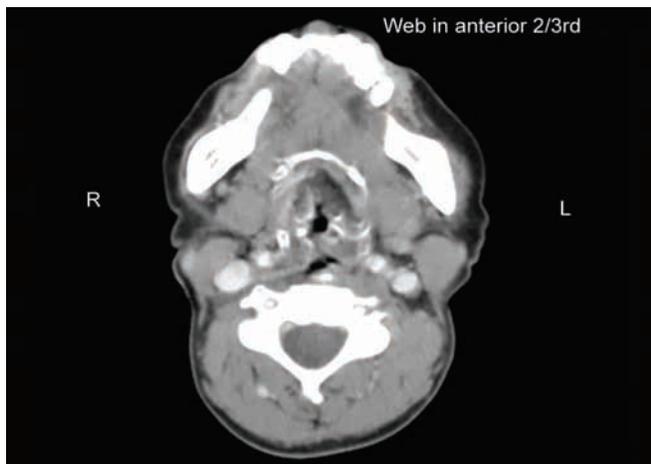


Fig. 1: Computed tomography scan showing web in anterior 2/3rd of vocal cord

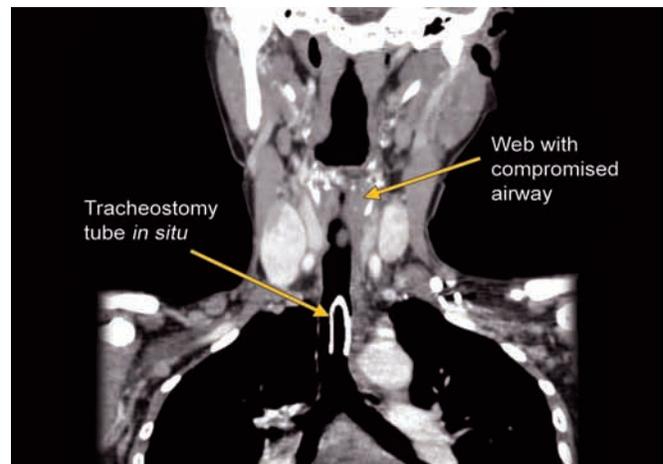
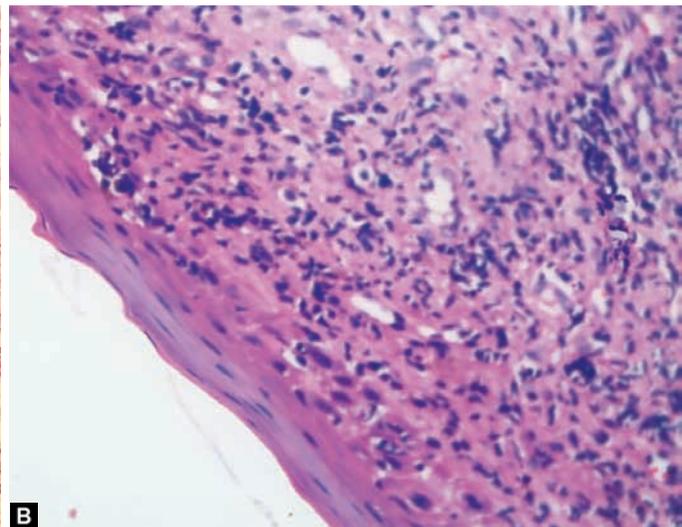
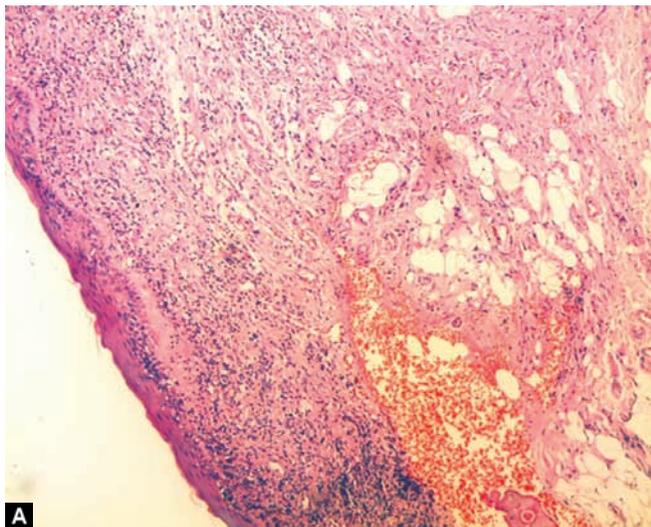


Fig. 2: Computed tomography scan showing tracheostomy tube *in situ* with compromised airway



Figs 3A and B: Fibromuscular tissue covered by stratified squamous epithelium exhibiting parakeratosis, irregular acanthosis. There is dense subepithelial infiltrates of lymphocytes with focal lymphoid follicles and exocytosis of lymphocytes. There is basal cell vacuolation, few colloid bodies and mild pigment incontinence

a carbon dioxide laser excision for the same. Thereafter, she was decannulated and tolerated the procedure well.

On subsequent follow-up at 1 year, there was no recurrence or residual of the lesions (Fig. 4).

DISCUSSION

Lichen planus is an uncommon disorder of unknown cause that most commonly affects middle-aged females which heal by scarring. Lichen planus may affect the skin, oral cavity, genitalia, scalp, nails, esophagus or any mucosal lined surface. However laryngeal involvement is extremely rare.³

Proposed pathophysiology of LP is an immune-mediated reaction against an exogenous or endogenous antigen which leads to the onset of the disease.

Potential triggers for the immune-mediated response include exposure to infections, trauma, drugs and contact allergens. However, a causative role for any of these factors in oral LP is still controversial. Various



Fig. 4: Postoperative fiberoptic image after 1 year

autoimmune diseases and viral infections like hepatitis C, herpes viruses, HIV have been associated to lichen planus, however, none of the correlations have been proved.^{3,4}

It is an inflammatory lesion wherein the lesions have a tendency to heal with scarring. Mucosal, cutaneous and coexistent mucocutaneous lesions have an equal preponderance in the body. Usually the lesions heal in about 18 months in majority of patients, however, lesions involving the oral cavity, genitals and other mucosal sites take longer time to heal.¹

It has been earlier well-proposed that both oral and laryngeal lichen planus affect mucous membranes and also behave clinically similar and therefore have been clubbed together as mucosal lichen planus.⁴

Mucosal lichen planus may have a varied presentation. oral lichen planus usually manifest as pain, burning and soreness. It is marked by involvement of buccal mucosa with characteristic white or gray streaks forming a linear or reticular pattern on a red purple background (Wickham striae).⁵

Clinical presentation of lichen planus depends on the anatomical site involved and usually multiple sites are involved. Anogenital lesions present with pruritis and esophageal lesions are known to cause dysphagia. Very rarely if it involves the larynx, it presents as hoarseness of voice, as seen in the present case.²

The most common differential diagnosis of mucosal lichen planus includes leukoplakia, fungal infection, carcinoma *in situ*, carcinoma and tuberculosis.⁶

Chronic mucosal lesions have a rare tendency to transform into malignancy which has been reported in cases of oral lichen planus. The reported incidence of malignant transformation vary from 1.2 to 5.3%. However, no such transformation has been reported in cases of laryngeal lichen planus.⁷

Smoking, excessive alcohol consumption, and atrophic, ulcerative or erosive disease are potential risk factors for the above malignant transformation.⁸

Characteristic histopathological appearance of the lesion is the most important diagnostic tools. Characteristic histopathological findings include a band-like lymphocytic infiltrate at the dermoepidermal junction, with damage to the basal cell layer. Dermoepidermal junction may also demonstrate characteristic colloid or civatte bodies (degenerated keratinocytes). Lymphocyte and plasma cell infiltration is more prominent in cases of mucosal lichen planus.^{5,9}

Treatment of lichen planus includes removal of potential triggering factors and maintaining good hygiene. Corticosteroids remain the drug of choice for medical management of lichen planus. If the site is accessible topical corticosteroids are effective in majority of patients.¹⁰

Systemic corticosteroids may be used in conjunction to topical steroids or in cases when topical treatment fails, or in cases with erosive or extensive lesions.⁹

The usual dose is 0.5 to 1.0 mg/kg/day for 2 to 12 weeks, depending on treatment response. If steroids are contraindicated or the in steroid-dependent patients azathioprine may be prescribed. Tacrolimus, dapsone, antimalarials have been tried with varied success rates in refractory cases.¹¹

CONCLUSION

Lichen planus is a common, chronic mucocutaneous disease associated with a cell-mediated immunological dysfunction. It may affect various sites both mucosal and cutaneous with varied clinical manifestations. Although rare, laryngeal lichen planus should be considered in the differential diagnosis of chronic hoarseness. The preset clinical scenario of lichen planus involving oral, laryngeal and genital areas is to our knowledge the first case to be reported from this part of the subcontinent.

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