

# A Rare Case of Malignant Transformation of Oral Lichen Planus of the Mandible

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**Summary:** Oral lichen planus (OLP) is an immune-mediated mucocutaneous disease associated with an increased risk in oral squamous cell carcinoma (OSCC). Nearly all cases of malignant transformation have been reported in patients >40 years old. We report the case of a 37-year-old woman with a 5-year history of erosive OLP who presented with malignant transformation to OSCC. Delineating the margins of the disease was impossible at presentation given her OLP, and she was initially treated with concurrent chemoradiation therapy. She then developed a recurrence of the mandibular alveolar ridge. The patient was successfully treated with a composite resection including a segmental mandibulectomy, buccal mucosa resection, partial glossectomy, and ipsilateral neck dissection. This was reconstructed with a free fibula osteo-septo-cutaneous flap. Mandibular OSCC is a rare complication of OLP with few reports on effective reconstructive interventions. The case represents the youngest reported patient with mandibular OSCC arising in the context of OLP and highlights the utility of the free vascularized fibula graft in the treatment of these patients. (*Plast Reconstr Surg Glob Open* 2016;4:e1070; doi: 10.1097/GOX.0000000000001070; Published online 23 December 2016.)

Oral lichen planus (OLP) is T cell-mediated chronic inflammatory disease of the oral mucosa. In the erosive form, the lesions tend to be multifocal, ulcerative, and painful.<sup>1</sup> Patients with OLP have an increased risk of developing oral squamous cell carcinomas (OSCCs), with a reported malignant transformation rate between 0% and 5.3%.<sup>2</sup> Lesions in the tongue and buccal mucosa are most likely to undergo malignant transformation.<sup>3</sup> Malignant transformation of OLP in a patient under age 40 is exceedingly rare, although cases have been reported in patients as young as 17.<sup>4</sup> In this study, we present a 37-year-old patient who developed OSCC of the buccal mucosa and mandibular alveolar ridge in the setting of a 5-year history of erosive lichen planus. This was treated with a composite resection and reconstructed with a free osteo-septo-cutaneous fibula flap.

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## CASE REPORT

The patient is a 37-year-old woman with a history of erosive OLP diagnosed by biopsy in 2010. She presented for follow-up in 2015 with a 3-month history of increased ulceration, swelling, and pain in her right buccal mucosa. Examination revealed a 2.5-cm hard, irregular, friable, erosive mass on the right buccal mucosa with extension to the retromolar trigone and inferior gingivobuccal sulcus. A biopsy demonstrated invasive, moderately to poorly differentiated squamous cell carcinoma (SCC). Positron emission tomography/computed tomography (PET/CT) revealed a 5- × 3-cm primary tumor with enlarged, PET-avid level IB and II lymph nodes, consistent with metastatic disease.

The erosive and extensive nature of the OLP made defining the margins impossible. Given the potential morbidity of an extensive oral cavity resection, the patient elected to undergo a 7-week course of concurrent radiation (70 GY) and chemotherapy with cisplatin for her clinical stage IV OSCC (T3N2bM0). Maxillofacial examination and panoramic radiographs revealed loss of bone and supporting structures associated with teeth #30 and #31 and woody fibrosis of the neck, consistent with post-radiation changes. Extraction of teeth #30 and #31 was performed after 20 treatments of hyperbaric oxygen therapy. At this time, biopsies of the right lateral tongue and

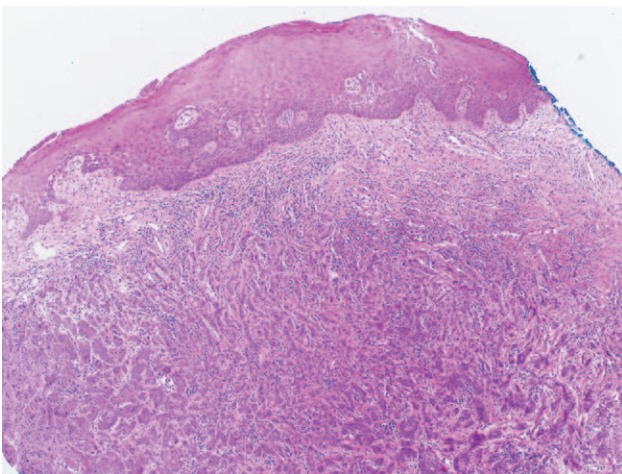
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gingiva were taken. Unfortunately, the gingival biopsies showed invasive, moderately to poorly differentiated SCC indicative of disease persistence (Fig. 1). Follow-up PET/CT demonstrated avidity in the mandible but no regional or metastatic disease.

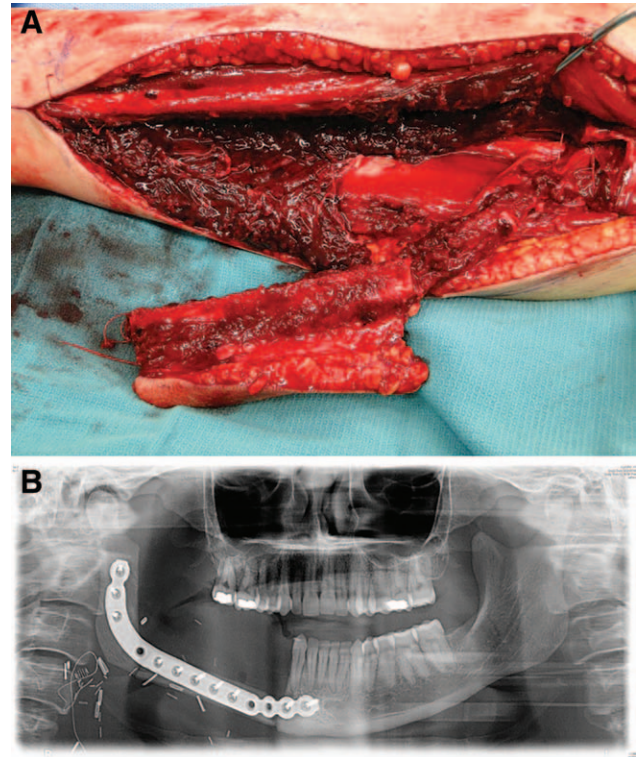
The patient subsequently underwent composite resection with right segmental mandibulectomy, partial glossectomy, resection of the buccal mucosa, and right selective neck dissection with a tracheostomy for airway protection. After completion of the extirpation, the patient was noted to have a 5.5-cm defect of the right mandibular body with a 4 × 8-cm intraoral mucosal defect. A left-side free osteo-septo-cutaneous fibula graft was used for reconstruction with microvascular anastomosis to the superior thyroid artery and the external jugular vein (Fig. 2). Surgical margins and sampled lymph nodes were negative for SCC. Final pathology was T1N0. The donor site was reconstructed using a split-thickness skin graft. Her postoperative course was uncomplicated, and she was discharged home on postoperative day 11. The patient was initially fed through a gastrostomy tube placed preoperatively. She was started on clear liquid diet on postoperative day 30. The patient has had appropriate wound healing at 6-week follow-up without complications (Fig. 3).

## DISCUSSION

Malignant transformation of OLP was first described by Koberg et al.<sup>5</sup> Since then, there have been numerous case reports and cohort studies associating OLP with OSCC, prompting the World Health organization to categorize OLP as a potentially malignant disorder.<sup>6</sup> Erosive and atrophic forms of OLP have the highest risk for malignant transformation.<sup>3</sup> Patients with OLP are advised to avoid additional risk factors such as tobacco and alcohol and are followed at minimum yearly.<sup>1</sup> However, it remains unclear to what extent OLP is an independent risk factor for the development of OSCC. The ability to draw definitive conclusions from the literature is limited by the inability to perform randomized control trials and inconsistency



**Fig. 1.** Biopsy demonstrating invasive SCC infiltrating submucosa (H&E, 100×).



**Fig. 2.** Mandibular reconstruction with free fibula osteo-septo-cutaneous flap. A, Fibula osteo-septo-cutaneous flap in situ after harvesting. B, Reconstruction plate (KLS martin) and fibula free flap as seen on postoperative orthopantomogram.

of inclusion criteria. Studies have been confounded by the presence of other risk factors, variable inclusion of oral lichenoid lesions, and lack of documentation of whether the cancer arose in the OLP or whether it was merely associated with it.

The pathogenesis of OLP and the mechanism for malignant transformation are unclear. OLP is thought to be a CD8+ T lymphocyte-mediated autoimmune disease affecting the basal keratinocytes. Alterations in the expression of a variety of factors including p53, COX-2, Smad3, Bmi1, CD133, and Myc-1 have been reported to be significant predictors of malignant transformation.<sup>7</sup> The prevailing theory proposes that neoplastic change occurs because of chronic stimulation by inflammatory mediators released from infiltrating T cells and subsequent oxidative stress.<sup>8</sup>

Mandibular extension is uncommon in OSCC developing from OLP with a review of the literature revealing fewer than 10 reported cases. Nearly all of these cases occurred in patients >50 years old; our patient represents the youngest reported case. The treatment of mandibular OSCC is complex because of the role of the mandible in mastication and speech and the need for satisfactory aesthetic outcomes. We recently described the versatility of the free vascularized fibula graft for mandibular reconstruction.<sup>9</sup> Based on the defect, an osseous, osteo-septo-cutaneous, or osteomyocutaneous flap can be used. The skin island can provide tissue for reconstruction of both external and oral cavity soft-tissue defects.<sup>10</sup> In cases of height



**Fig. 3.** Intraoral view of the reconstructed mandible demonstrating transformation of the intraoral skin island into mucosa as early as 6 weeks after surgery.

discrepancy, a “double-barrel” modification enables bridging of the defect and immediate osseointegrated implantation with improved outcomes. Finally, preparation of 3-dimensional models of fibula grafts before surgery and use of prefabricated plates and cutting systems guarantee the proper orientation and alignment of the osseous substructure in relation to the maxillary bone,<sup>10</sup> facilitating a functional dental rehabilitation. Other techniques for fibula reconstruction can result in osseous continuity of the mandible, but the position of the graft may preclude or

compromise the ability for dental rehabilitation to achieve both a functional and aesthetic result.

To our knowledge, this represents the youngest reported patient with mandibular OSCC arising from OLP. The case demonstrates the efficacy of mandibular reconstruction with a fibula osteo-septo-cutaneous flap in this patient population.

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