Lichen planus (LP) is an autoimmune inflammatory dermatosis of unknown origin that typically affects the skin, nails, and hair. Lichen planus can also involve mucosal surfaces of the mouth, conjunctiva, nose, larynx, esophagus, and anogenital area. Although LP affects about 1% of adults, the diagnosis and treatment of mucosal LP are frequently missed or delayed. The stratified squamous epithelium of the external auditory canal and the tympanic membranes can be affected, albeit rarely, leading to otorrhea, external auditory canal stenosis, tympanic membrane thickening, and conductive hearing loss. The prevalence of otic LP is difficult to estimate; indeed, only 1 patient. In the remainder, extraotic LP had been diagnosed for several years (usually 1-5 years) before patients noticed otic disease.

We identified 19 cases of otic LP. Table 1 provides the overall patient characteristics and sites of LP. Table 2 gives patient-specific data, including demographic characteristics, clinical findings, therapy and outcome, and duration of follow-up. The most common concerns were otorrhea and hearing loss (15 patients each); 11 of these patients presented with simultaneous otorrhea and hearing loss. Plugging (n = 6) and pruritus (n = 5) were also common concerns. The mean duration of symptoms before diagnosis was 4.0 years in 13 patients (range, 0.5-14 years). Five patients had otic LP only. In those with LP involving multiple sites, otic disease was the presenting manifestation in only 1 patient. In the remainder, extraotic LP had been diagnosed for several years (usually 1-5 years) before patients noticed otic disease.

**Report of Cases**

The study protocol was approved by the Mayo Clinic institutional review board. We reviewed the electronic medical records of patients with otic LP who presented to Mayo Clinic (Rochester, Minnesota) from January 1, 2001, through May 31, 2011. The institutional master diagnosis index database was queried using the keywords *lichen planus, otitis, ear canal, otic lichen planus, and auditory canal stenosis*. Relevant clinical information, including sex, race, age at diagnosis, ear symptoms, physical examination findings, histopathologic examination findings, previous diagnoses, treatment, and response to therapy, was evaluated. Lichen planus at other anatomic sites was also documented.
Examination of the ears showed 11 patients with bilateral disease (58%) and 8 with unilateral involvement (the right ear in 4 patients). Otoscopic examination findings are provided in Table 3. Mild erythema, painless induration and stenosis of the external auditory canal, and thickening of the tympanic membranes were the most common abnormalities (Figure).

Histopathologic examination was performed in 12 patients. Of these, 3 patients underwent a skin biopsy of the external auditory canal only (2 showed granulation tissue; 1 showed changes of focal chronically inflamed fibrous tissue) and 9 underwent biopsies of extraotic sites (ie, the lip, buccal mucosa, esophagus, vulva, and skin). Light microscopy was diagnostic for LP in 8 patients, 6 of whom also had direct immunofluorescence results that showed the characteristic finding of a strong, shaggy deposition of fibrinogen along the basement membrane zone and patchy staining of the subepithelial mucosa. Notably, this classic pattern was found only at extraotic sites. In the remaining 4 patients without classic biopsy findings and in the 7 patients who did not undergo biopsy, otic LP was diagnosed on the basis of clinical findings alone (ie, classic symptoms of ear discharge and hearing loss, characteristic otoscopic examination findings, and response to treatment). Before the final diagnosis of otic LP was established, 5 patients had received other diagnoses that included isolated ear canal stenosis and chronic inflammation, chronic myringitis, chronic external otitis, and chronic serous otitis.

Seventeen patients received therapy with topical tacrolimus. In our anecdotal experience, patients with oral LP respond better to tacrolimus therapy than to topical corticosteroid therapy; by extension, we usually initiate treatment with topical tacrolimus. The other 2 patients received topical clofetrol propionate or a combination therapy of otic ciprofloxacin hydrochloride and dexamethasone.

The mean duration of follow-up was 4.9 years (range, 0.5-11 years) from the initial diagnosis of LP, and the mean number of follow-up dermatology and otolaryngology visits was 13.8 (range, 2-50 visits). Three patients with isolated otic LP were not referred to a dermatologist, and their LP was managed primarily by otolaryngologists. Gastroenterologists and gynecologists also evaluated the condition of a small number of patients with esophageal and genital disease, respectively.

Systemic therapy was not necessary to control otic LP, but 3 patients received systemic agents for the treatment of severe and persistent extraotic LP; such treatments included methotrexate sodium, hydroxychloroquine sulfate, prednisone, dapsone, mycophenolate mofetil hydrochloride, azathioprine, and systemic tacrolimus. Furthermore, 1 patient with Sjögren syndrome had been intolerant of systemic tacrolimus because of pruritus. This patient had severe LP affecting the ear, oral cavity, esophagus, and genital area and reported remarkable relief with rituximab therapy that had been prescribed primarily for the management of Sjögren syndrome.

Our cohort indicated that the most common otic complications were hearing loss (usually bilateral, with a conductive or mixed pattern) and external auditory canal stenosis. Because of the severity of hearing loss, 4 patients received bilateral hearing aids. Of these 4 patients, one underwent right stapedectomy because of otosclerosis and another underwent excision and reconstruction of a left external auditory canal stenosis years before the diagnosis of otic LP was established. Extraotic complications included recurrent esophageal strictures (causing dysphagia and odynophagia and necessitating multiple esophageal dilations), lacrimal duct stenosis, erosive mucositis, vaginal stenosis, and fusion of the external genitalia (causing dyspareunia and the inability to have sexual intercourse).

### Discussion

The major findings of our study were as follows: (1) otic LP had a nonspecific presentation; (2) otic LP was most commonly seen in patients with LP affecting other sites, but it also presented as an isolated condition; (3) the female to male ratio was 3.75:1; (4) dedicated histopathologic examination of the external auditory canal was not necessary to establish the diagnosis; (5) topical tacrolimus was a reasonable therapeutic strategy, with an overall good response within several months; (6) hearing loss and external auditory canal stenosis were among the usual and expected complications of otic LP; and (7) systemic treatment with rituximab may be considered for disseminated disease.

The study has limitations, mainly owing to its retrospective nature. First, the true incidence of otic LP may be underestimated because all patients with otic symptoms, including otorhea and hearing loss, theoretically would require an examination to exclude otic LP. Understandably, this is impossible in a retrospective case series. Also, some patients with more generalized cutaneous disease may have been missed when auditory examinations were not performed. Since the initial observation by Warin et al in 1948 of LP affecting the tympanic membranes, otic LP has been reported infrequently in the literature. To our knowledge, our cohort represents the largest series on otic LP in the English-language medical literature. Similar to the findings of Martin et al, hearing loss was the most common symptom, affecting 15 patients.
In addition, painless otorrhea, likely attributable to continuous inflammation of the external canal, was the present- ing concern as frequently as hearing loss.

As previously reported,5,8 physical examination disclosed mild erythema and strictures of the external auditory canal in more than half the patients (11 of 19). Of note, exci-
The diagnosis of otic LP was determined by histopathologic examination of extraotic sites or solely on the basis of clinical observations. In 8 patients, light microscopy findings were consistent with LP; 6 also showed the classic shaggy fibrinogen deposition along the basement membrane zone on direct immunofluorescence. When the clinical suspicion for otic LP is high or the diagnosis is supported by biopsy-proven LP at other sites, prompt referral and early treatment is recommended. In addition, we recommend skin biopsy of the external auditory canal to exclude an alternative diagnosis only if the LP does not respond to a therapeutic trial with topical tacrolimus.

We have used custom-compounded tacrolimus, 0.1%, in either mineral oil or the combination of neomycin sulfate, polymyxin B sulfate, and hydrocortisone for successful treatment of this condition. Although topical neomycin can trigger classic contact dermatitis reactions and may be inappropriate for long-term treatment of susceptible individuals, we did not encounter this reaction in our cohort. In our experience, twice-daily application showed objective improvement by the time of the first follow-up visit, 1 month after initiating treatment. These results were consistent with those reported by Harth et al, who demonstrated that topical tacrolimus applied to the external auditory canal markedly improved symptoms in up to 85% of patients with chronic external otitis. Our findings also are supported by Djallil and Memar, who showed resolution of refractory pruritic external auditory canals in more than 90% of patients who had not responded to conventional treatment.

In our cohort, one patient attained sustained relief with rituximab administered for the treatment of Sjögren syndrome. A published case report similarly showed that 1-month treatment with intravenous rituximab resulted in a striking improvement of symptoms—mainly dysphagia due to esophageal LP—for up to 10 months. Cyclosporine and oral tacrolimus also have been used successfully in patients with mucosal LP. These provocative data suggest that systemic treatment may be an appealing alternative to topical medications, especially in patients with recalcitrant and disseminated disease.

In conclusion, otic LP should be in the differential diagnosis of persistent otorrhea, ear canal inflammation, and hearing loss, particularly when symptoms are refractory to stan-
standard treatment and if there is evidence of extraotic LP. For the initial treatment, we recommend topical tacrolimus, 0.1%, in either mineral oil or topical corticosteroid devoid of neomycin, applied at least twice daily, with regular follow-up care by a dermatologist and an otolaryngologist. For recalcitrant disease or widespread involvement, a multidisciplinary approach with assessment by specialists may be warranted before initiating systemic treatment (eg, rituximab), at least until the efficacy and safety have been validated by prospective, placebo-controlled studies. Finally, appropriate and timely referrals to a dermatologist should be made for patients with suspected mucosal LP.

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