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Case Report

Lymphoepithelioma- Like Carcinoma of the Skin Treated with Mohs Micrographic Surgery: Case Report and Review of Literature

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ABSTRACT

Lymphoepithelioma-like carcinoma of the skin (LELCS) is a rare cutaneous neoplasm that histologically resembles nasopharyngeal lymphoepithelioma linked to Epstein-Barr virus (EBV)1. First described in 1988, LELCS presents as a red or flesh-colored nodule or plaque, on the head and neck, in middle-aged to elderly patients2. It is characterized by aggregates of malignant undifferentiated cells in the dermis surrounded by reactive lymphoplasmacytic infiltrate1,2. Recurrence and metastases are probable if inadequately treated. Specific guidelines for treatment have not been established. There are a few reported cases treated with Mohs micrographic surgery (MMS). We report a case of LELCS treated with MMS and review the relevant literature.

Keywords: Lymphoepithelioma like carcinoma of the skin; Mohs surgery; Wide local excision; Skin cancer; Oncology; Surgery

Case Report

A 79-year-old Caucasian female presented with a 3-month history of enlarging red nodule on the temple. Clinically, a solitary, ill-defined, deep dermal nodule measuring 1.5 cm x 1 cm was noted (**Figure 1a**). No regional lymphadenopathy or nasopharyngeal abnormalities were noted. Excision performed for a presumed inflamed cyst revealed infiltrating dermal tumor with atypical histiocytoid proliferation surrounded by dense lymphocytic infiltrate on histopathology (**Fig.1b, 1c**). Immunohistochemical staining with pan-cytokeratin AE1/AE3, cytokeratin 5/6, and p63 were positive, suggesting epithelial, cutaneous origin. Cytokeratin 7 and cytokeratin 20 were negative, ruling out neuroendocrine tumors. EBV mRNA was not detected. Based on microscopy, and immunohistochemistry (IHC), a diagnosis of LELCS was made. Patient underwent MMS, and tumor-free margins were achieved after two stages. No evidence of disease (NED) was noted at 15 months postoperatively.

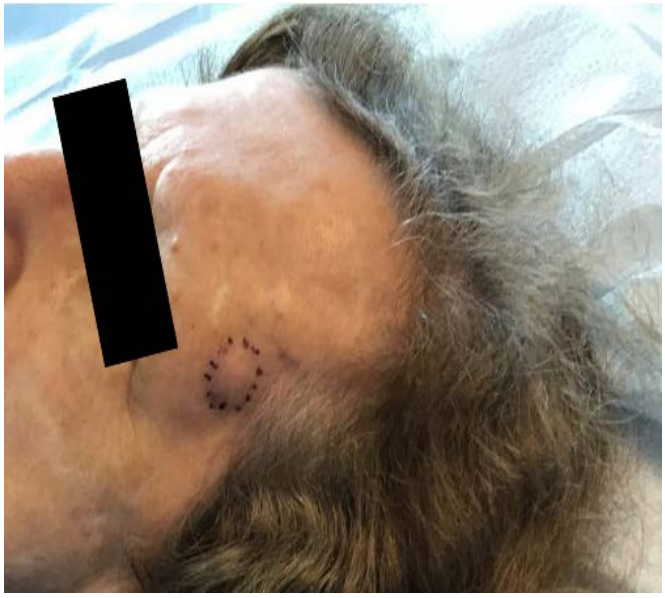

Figure 1a. Solitary, ill-defined, deep dermal nodule measuring 1.5 cm x 1 cm with overlying erythema.

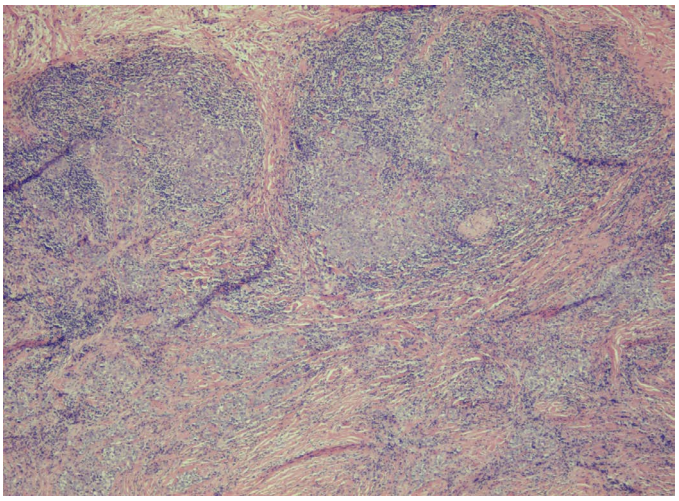

Figure 1b. Infiltrating dermal tumor with a histiocytoid proliferation and dense peripheral lymphocytic infiltrate (H&E, 4x original magnification)

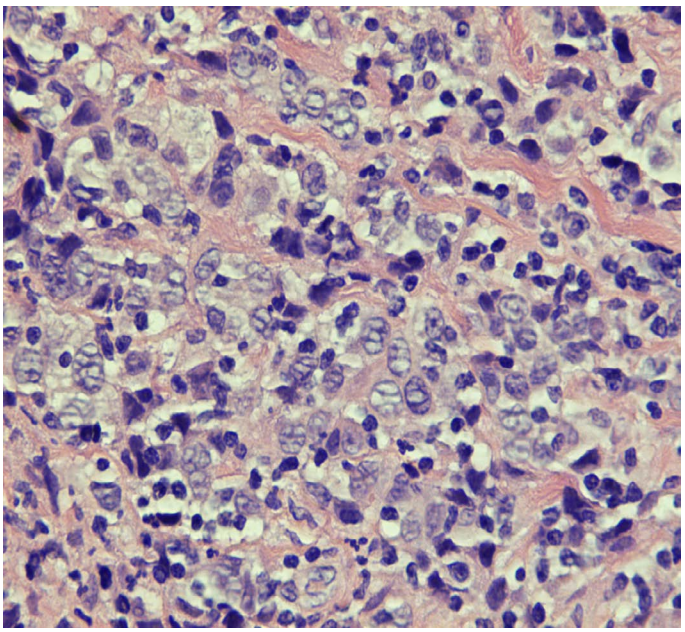

Figure 1c. A nest of neoplastic cells with nuclear pleomorphism, and dense lymphoplasmacytic infiltrate at the periphery of neoplastic cells (H&E, 40x original magnification)

Discussion

LELCS is a rare cutaneous neoplasm that presents as a flesh-colored or red nodule, on the head and neck of middle-aged and elderly, with equal incidence in men and women[3,4]. LELCS is histologically composed of poorly differentiated tumor cells in the dermis surrounded by dense lymphoplasmacytic infiltrate, similar to poorly differentiated nasopharyngeal carcinoma associated with EBV[2,3]. IHC differentiates LELCS from Merkel cell carcinoma, lymphoma, and pseudolymphoma[2,5]. Tumor cells in LELCS are positive for high-molecular-weight cytokeratins and epithelial membrane antigens, indicating epithelial origin[2,3]. Expression of EBV nucleic acid is variable in cutaneous neoplasms[2]. Clinical characteristics, management of 91 cases of LELCS reported in the literature are summarized in (**Table 1**). Briefly, LELCS affected the elderly, with a mean age of 73 years (39-97 years). Male to female ratio was 1.5:1. Locations involved include face (87.9%), scalp (5.5%), arm/shoulder (2.2%), trunk (2.2%), and penis (2.2%). Most common presentation was a solitary nodule or papule. MMS was performed in 8 of 91 patients (8.8%). WLE was performed in 64 of 91 patients (70.3%), treatment in 17 of 91 (18.6%) cases was unknown. Electrocautery with a deep shave, slow mohs[6-11] and WLE with en face permanent sections were done in three cases. No evidence of recurrence or metastasis was reported in cases treated with MMS. Eight out of 63 cases (11.1%) treated with WLE displayed local recurrence, needed adjuvant therapy or retreatment. Two patients died from metastases. Standard treatment of LELCS includes WLE or MMS[1,2]. MMS provides greater deep-margin assessment as the deep dermal nature of the tumor makes it difficult to diagnose its full extent. Clinical features of patients treated with MMS are summarized in (**Table 2**). Close clinical follow-up is recommended. Management should include an otolaryngological exam, lymph node assessment. Distant metastases, although rare has been reported[2]. LELCS is radiosensitive; radiation is reserved for unresectable tumors, and adjuvant therapy[2]. In conclusion, LELCS is a rare cutaneous neoplasm with a high rate of recurrence and MMS with complete margin assessment reduces recurrence.

Table 1: Clinical and demographic features of the 91 cases of lymphoepithelioma- like carcinoma of the skin reported between1998-2022.

Category	Data
Age (n=91)	39-97 (mean, 73 years)
Sex (n=91; M:F)	1.5:1
Location (n=91)	Face: 80 (87.9%); scalp 5 (5.5%); arm/shoulder 2 (2.2%); trunk 2 (2.2%); penis 2 (2.2%)
Morphology at presentation (n=91)	Solitary nodule
Management (n=91)	Local excision: 64 (70.3%); (1 out of 64 had en face permanent section margin control); Mohs micrographic surgery 8 (8.8%); Slow Mohs 1(1.1%); Electrocautery with deep shave 1 (1.1%); Unknown 17 (16.4%)
Outcome (n=91)	NED with MMS: 8/8 (100%). NED with surgical excision: 32/64 (50%) cases. Recurrence was noted in 8/64 cases (12.5%) Follow-up N/A: 9/64 cases Metastases to distant organs: 5 Death from metastasis:2 Died due to unrelated causes:5

NED - No evidence of disease

MMS- Mohs micrographic surgery

N/A- Not available

Table 2. Clinical features of patients treated with MMS and total microscopic margin control.

Reference	Age (years), Sex	Presentation	Location	Management	Follow up
Glaich et al. 2006[5]	97, F	Nodule	Cheek	MMS	NED, at 6 months
	88, F	Nodule	Arm	Wide excision with en face permanent sectioning	NED at 10 months
Robins et al. 1995[4]	74, F	Nodule	Cheek	MMS	NED, unknown duration
Dudley et al. 1998[7]	81, M	Nodule	Mandible	MMS	NED, unknown duration
Dozier et al. 1995[8]	91, F	Pearly papule	Dorsal nose	Electrodessication and curettage; MMS at recurrence	NED, at 20 months
Jimenez et al. 1995[9]	68, M	Plaque	Nasal ala	MMS	NED at 12 months
Lyle et al. 2008[10]	68, F	Nodule	Forehead	MMS	NED at 5 months
Wang et al. 2014[11]	60, M	Nodule	Cheek	MMS	NED, unknown duration
Our case	78, F	Nodule	Temple	MMS	NED at 6 months
Tzanidakis et al. 2015[6]	F,79	Temple	Nodule	Slow Mohs	NED, unknown duration

NED - No evidence of disease

MMS- Mohs micrographic surgery

N/A- Not available

M- Male F-Female

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Reprint requests: Sujitha Yadlapati

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