

# Trichilemmal Carcinoma In a Lung Transplant Recipient

M Hamodat, S Ryan, P Wadden

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## Abstract

Trichilemmal carcinoma is a rarely diagnosed cutaneous adnexal neoplasm. It is derived from or differentiates towards the outer root sheath of the hair follicle and is the malignant counterpart of trichilemmoma. Here we report a case of TLCA that developed in a 68 year old male lung transplant recipient who was on immunosuppressive therapy. This patient has a long history of multiple skin tumors including basal cell carcinoma. Reports of TLCA in the literature are limited. In particular, very few cases of TLCA in immunosuppressed patients have been reported. We will review the current literature in the area and discuss the need for specific pathological analysis of all excised lesions from this patient population.

## INTRODUCTION

Trichilemmal carcinoma is a rare cutaneous adnexal malignant tumor deriving from the outer root sheath of hair follicles (trichilemma); therefore it represents the malignant counterpart of trichilemmoma.<sup>1</sup> Headington first proposed the term trichilemmal carcinoma (TLCA) for a "histologically invasive, cytologically atypical clear cell neoplasm of adnexal keratinocytes which is in continuity with the epidermis and/or follicular epithelium".<sup>2</sup>

TLCA usually affects elderly patients in their eight decade and presents as a vegetating, often ulcerated or crusted nodule or plaque located preferentially on sun-exposed areas, mostly the head, neck and dorsum of the hand.<sup>3,4,5,6</sup>

Here we report a case of TLCA that developed in an immune-suppressed lung transplant recipient with a history of multiple skin cancers to which these patients are prone. TLCA has been only rarely been reported in this clinical setting. We will also review the current literature in this area.

## CASE REPORT

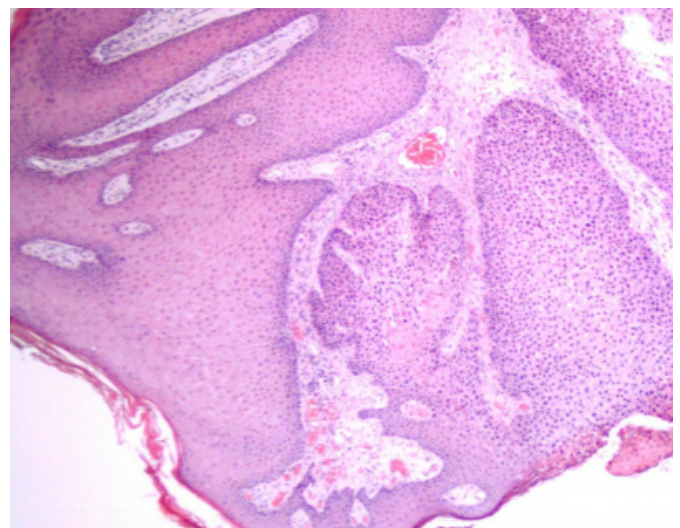
We report the case of cutaneous TLCA excised from the neck of a 68 year old male who was on long-term immunosuppressive therapy following a lung transplant six years ago. Multiple skin cancers including basal cell carcinoma had been diagnosed during this time. The patient's most recent clinically significant cancer was a superficially invasive squamous cell carcinoma of the vocal cord.

Grossly, the skin ellipse showed a 1.5cm tan-brown papular

lesion with surface crusting. Microscopically an exo-/endophytic tumor was formed of multiple epithelioid lobules emanating from the surface and expanded upper follicular zones. There was superficial erosion and lateral molding by a basal collarette of reactive squamous epithelium

## Figure 1

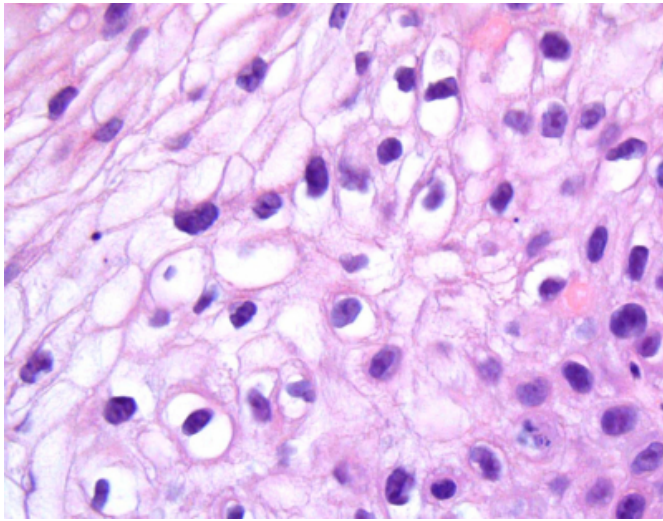
Figure 1: Hematoxyline & Eosin, original magnification X 40. Apolypoid lesion that has surface erosion and consist of multiple endophytic lobules of squamous epithelium projecting in deeper dermis



The lobules showed peripheral palisading, focal eosinophilic basement membrane mantle and spotty central micro pustules or trichilemmal type of keratinization but most lobules had solid centers.

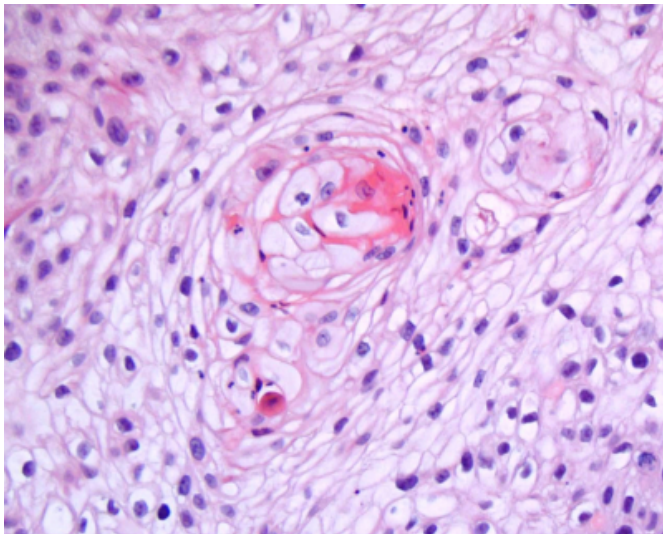
**Figure 2**

Figure 2: Hematoxyline & Eosin, original magnification X 40. Zonal arrangement of cells with prominent clear cytoplasm



**Figure 3**

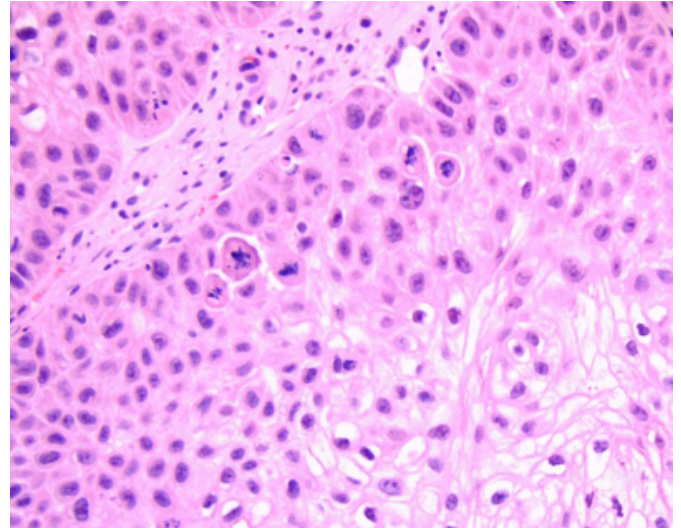
Figure 3: Hematoxyline & Eosin, original magnification x 40. Some centers show abrupt keratinization.



Cytologically, peripheral columnar cells and central polygonal cells had prominent cleared cytoplasm. The nuclei were round; not indented like sebaceocytes. The tumor base lacked any inflammatory lichenoid infiltrate typical of keratoacanthoma. The presence of cytologic atypia, many mitotic figures, apoptotic necrosis and focally infiltrative leading edge indicated malignant features.

**Figure 4**

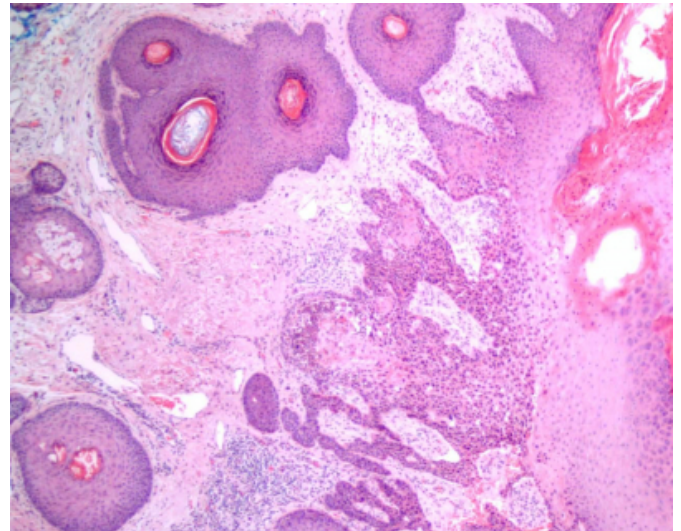
Figure 4: Hematoxyline & Eosin, original magnification X 40. Cytologically it is malignant characterized by pleomorphism, apoptotic necrosis and brisk mitosis but morphology in keeping with TLCA, an appendage tumor recapitulating the outer root sheath of hair follicle (tricholemmal sheath).



The tumor supporting stroma was fibromyxoid with prominent vascularity was in situ and microinvasive component.

**Figure 5**

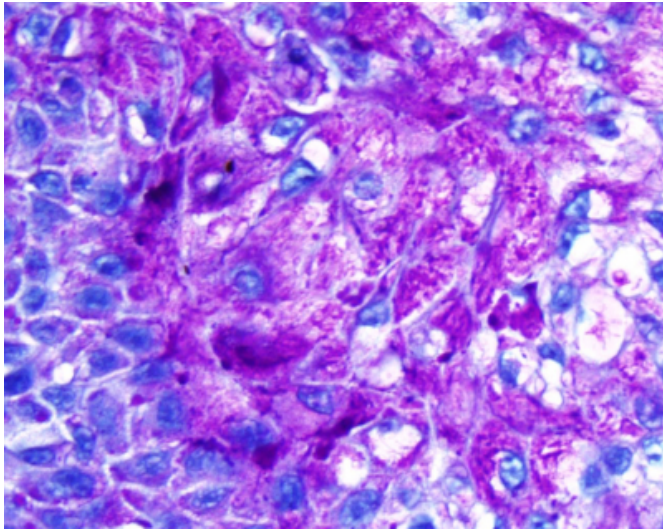
Figure 5: Hematoxyline & Eosin, original magnification X 40, In situ and microinvasive component



The background skin showed solar damage with confluent solar elastosis and telengectasias. PAS shows a high glycogen content in the lobules especially in the centre.

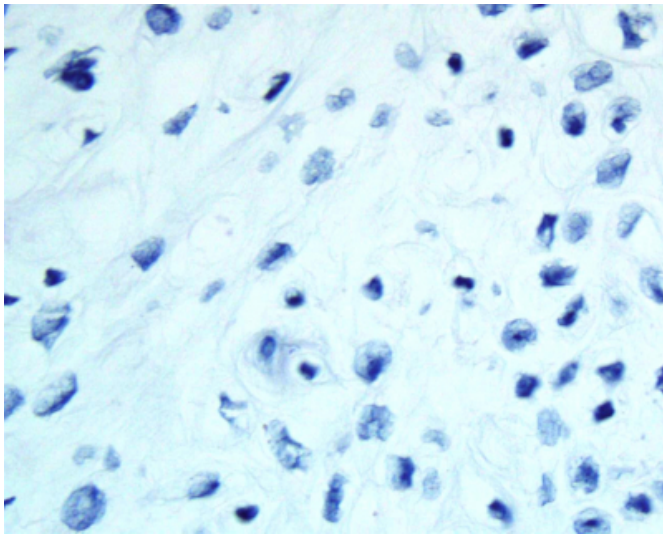
**Figure 6**

Figure 6: PAS and show a high glycogen content in the lobules especially in the centre, original magnification X 40.



**Figure 7**

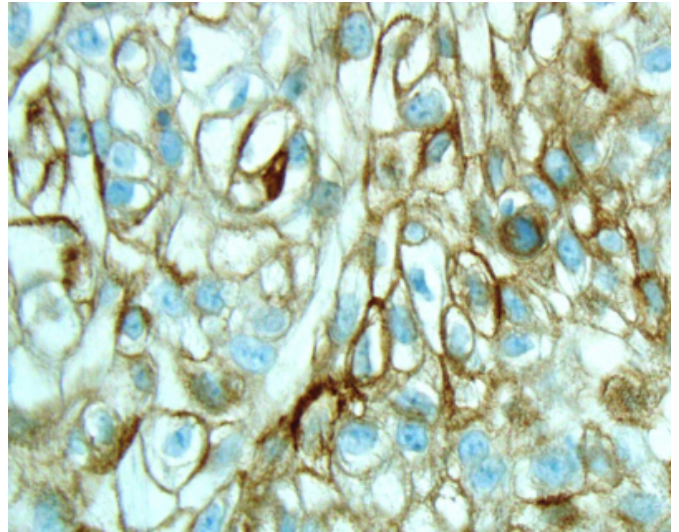
Figure 7: PAS – D shows a high glycogen content in the lobules especially in the centre, original magnification X 40.



EMA shows membrane staining in the central zone of the lobules.

**Figure 8**

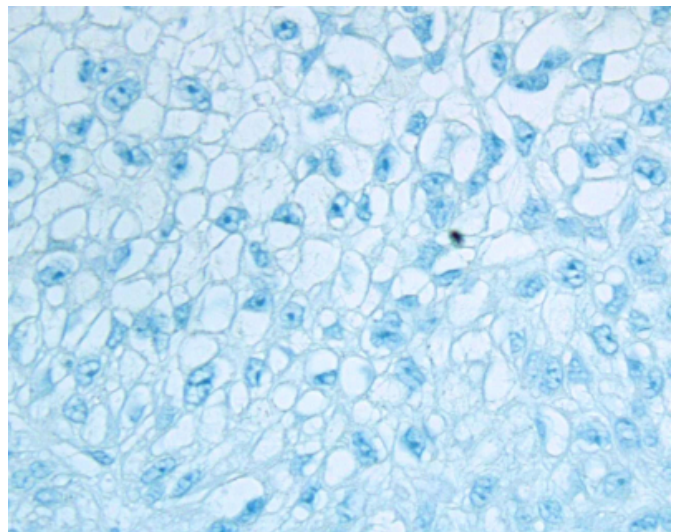
Figure 8: EMA shows membrane staining in the central zone of the lobules .original magnification X 40.



CEA is negative. Figure not shown. HPV stains are negative.

**Figure 9**

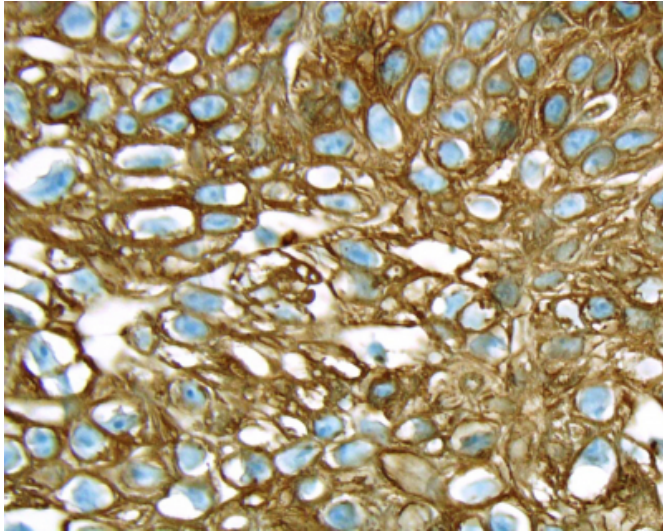
Figure 9: Negative HPV stain, original magnification X40.



CK5 is positive.

**Figure 10**

Figure 10: Positive ck5/6, original magnification X 40.



The resection margins of this carcinoma were free from evidence of TLCA thus no further excision was required. Healing of the excision in this patient was uneventful and no recurrence or metastasis of this tumor has occurred to date.

**DISCUSSION**

The etiology of trichilemmal carcinoma is unknown. Often the only recognizable symptom is the presence of an unusual, tan or flesh-colored spot on the skin. It has, however, been well documented that transplant recipients are at an increased risk to develop skin cancers, with squamous cell carcinoma being the most common.<sup>7</sup> Benign and malignant adnexal tumors develop more frequently in transplant recipients as compared to immunocompetent patients. TLCA however does not seem to follow this trend. It appears from our literature review that TLCA in transplant recipients has only been reported in three other cases.<sup>58</sup> However, the actual incidence of TLCA maybe underestimated because this tumor may be pathologically misdiagnosed as clear-cell squamous (or basal) cell carcinoma, sebaceous carcinoma or clear-cell Bowen's

disease.<sup>3</sup> Grossly, it simulates squamous cell carcinoma. Complete surgical excision is curative.

Despite the locally aggressive growth, TLCA is at a very low risk for recurrence or metastases, no reports of recurrence or metastases were observed in any previous cases reported in the literatures.

The etiology of TLCA remains unknown. Since TLCA develops preferentially on sun-exposed areas it is likely that ultraviolet radiation plays a role. Additional possible contributing factors in transplant recipient patients may include immunosuppressive treatment and HPV infection.<sup>1</sup> in our case, however, no evidence of HPV infection by morphology or immunohistochemistry was found.

In transplant recipient patients, TLCA and other cutaneous malignancies may initially appears benign thus underscoring the need for careful clinical evaluation with pathological analysis of all excised lesions from this patient population <sup>17</sup>.

**References**

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**Author Information**

**Mowafak Hamodat, MB.CH. MSc. FRCPC**

Eastern Health of St.John's, Memorial University of Newfoundland

**Suzanne C. Ryan, BSc**

Eastern Health of St.John's, Memorial University of Newfoundland

**Patricia M. Wadden, MD, FRCPC**

Eastern Health of St.John's, Memorial University of Newfoundland