
A Case of Desmoplastic Trichilemmoma of the Lip Treated with Mohs Surgery

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BACKGROUND. Desmoplastic trichilemmoma is a rare pseudo-malignant variant of trichilemmoma. It generally presents as a small papule on the face and is often clinically misdiagnosed as a basal cell carcinoma or verruca vulgaris. It is histologically similar to a trichilemmoma, but has a central area of desmoplasia that can mimic an invasive carcinoma.

OBJECTIVE. The objective was to report a case of desmoplastic trichilemmoma of the lower lip that was treated with Mohs micrographic surgery.

METHODS. A case is reported and the literature is reviewed.

ERIC SCHWEIGER, MD, CANDACE THORNTON SPANN, MD, JEFFREY M. WEINBERG, MD, AND BONNIE ROSS, MD HAVE INDICATED NO SIGNIFICANT INTEREST WITH COMMERCIAL SUPPORTERS.

TRICHILEMMOMA IS a benign neoplasm from the outer sheath of the pilosebaceous follicle, first described by Headington and French in 1962.¹ In 1990, Hunt et al.² described a variant of the neoplasm, desmoplastic trichilemmoma, which is characterized by a central zone of desmoplasia, which may mimic an invasive carcinoma. The clinical appearance of this tumor is often nonspecific, requiring histologic examination for diagnosis. We report a case of desmoplastic trichilemmoma of the lip that was treated with Mohs micrographic surgery.

Case Report

A 66-year-old Hispanic man presented with a 1-year history of a small tumor on his lower lip. The lesion reportedly bled often when he shaved. He denied any antecedent trauma, radiation, excessive sun exposure, cigarette smoking, or use of chewing tobacco. He had no personal or family history of skin cancers. On physical examination, the patient had a 5-mm flesh colored papule, with some central erosion, on his right

RESULTS. The patient underwent Mohs micrographic surgery for removal of the neoplasm. Six months after the procedure, the patient remained tumor free.

CONCLUSIONS. Although desmoplastic trichilemmoma is a benign neoplasm, it is often histologically confused with basal cell carcinoma and malignant trichilemmoma. Desmoplastic trichilemmoma is also most frequently located on the face. Considering these factors, Mohs micrographic surgery appears to represent an excellent choice for removal of these tumors to achieve clear margins and a good cosmetic result.

lower lip (Figure 1). A shave biopsy of the lesion was obtained.

Histologic examination revealed an exoendophytic epithelial neoplasm characterized by bulbous aggregates of small squamous cells emanating from a slightly papillated epidermis (Figure 2). Some of the cells had pale-staining cytoplasm. There was no evidence of cytologic atypia, but there were scattered mitotic figures. At the periphery, the epithelial aggregates had a smooth, rounded border. At the center, where the lesion was focally ulcerated, there were irregular anastomosing aggregates of epithelial cells admixed with a

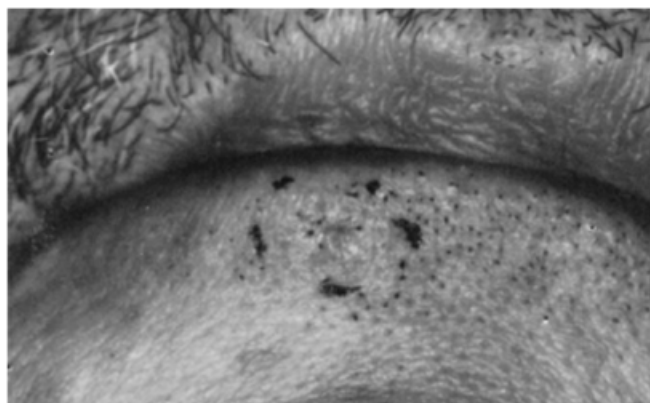


Figure 1. Desmoplastic trichilemmoma of the lower lip.

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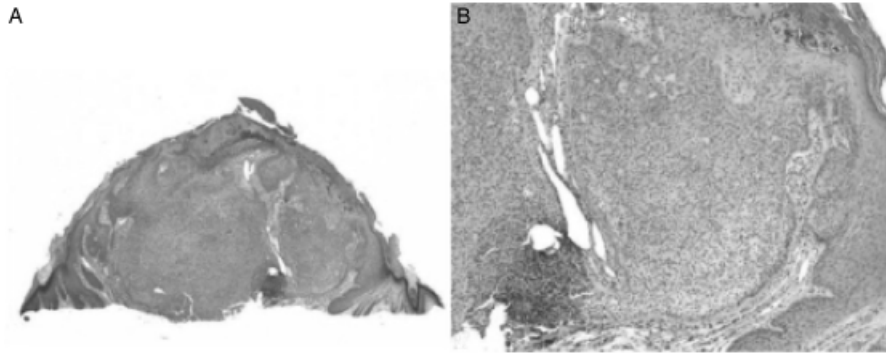


Figure 2. Exoendophytic epithelial neoplasm characterized by bulbous aggregates of small squamous cells emanating from a slightly papillated epidermis (hematoxylin and eosin; A, magnification $\times 100$; B, magnification $\times 400$).

fibromyxomatous and collagenous stroma (Figure 3). In this area, there was an infiltrate of lymphocytes and neutrophils. The diagnosis was believed to be consistent with desmoplastic trichilemmoma.

Because the lesion extended to the base of the specimen, complete removal of the tumor was recommended. The patient then underwent Mohs micrographic surgery to remove the lesion. At the time of Mohs micrographic surgery, one layer was required to clear the tumor. The resulting wound measured 0.7 cm and was closed primarily (Figure 4). The surgery was tolerated well with no complications. At follow-up 6 months after the procedure, the patient had not experienced a recurrence of the neoplasm.

Discussion

Desmoplastic trichilemmoma is a rare cutaneous variant of trichilemmoma. This neoplasm most commonly affects individuals after their fifth decade of life, but there have been reports of desmoplastic trichilemmoma in patients as young as age 8.^{2,3} The mean age of occurrence of desmoplastic trichilemmoma in one study was 64 years.² Clinically, the lesion is usually characterized as a dome-shaped papule with a smooth or keratotic surface, occasionally with pearly borders. Desmoplastic trichilemmomas are usually 5 to 7 mm in size and rarely exceed 1 cm². The most common location for this lesion is on the face. Tellechea et al.³ noted that four of seven patients with desmoplastic trichilemmoma they studied had the tumor located on the lip.³ Other common facial locations for the lesion include the nose, chin, cheek, and forehead.^{2,3} There have also been a few case reports report of desmoplastic trichilemmoma occurring on the eyelid.^{4,5} In addition to the face, desmoplastic trichilemmoma has also been found on the scalp, neck, chest, and vulva.² Desmoplastic trichilemmoma has also been documented occurring within a nevus sebaceous^{2,6} and arising in association with a basal cell carcinoma.⁷

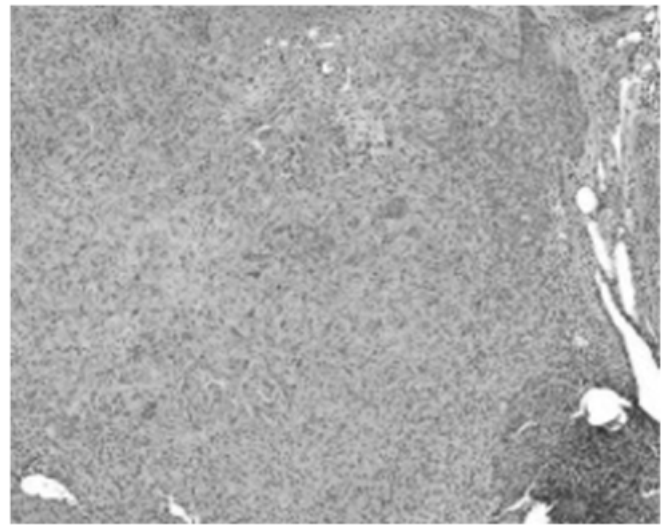


Figure 3. At the center, where the lesion was focally ulcerated, there were irregular anastomosing aggregates of epithelial cells admixed with a fibromyxomatous and collagenous stroma (hematoxylin and eosin; magnification $\times 400$).

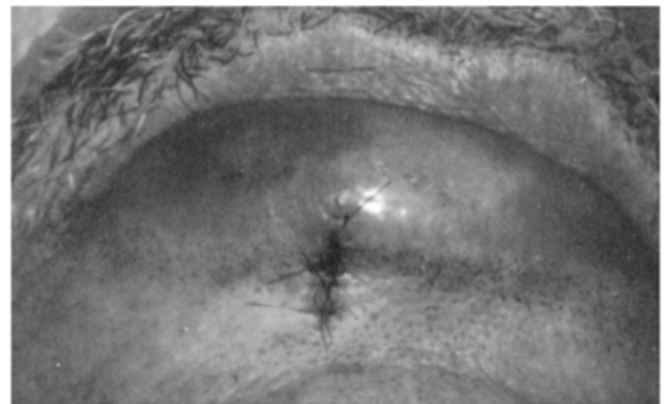


Figure 4. Primary closure after Mohs micrographic surgery.

Because of its nonspecific clinical appearance, desmoplastic trichilemmoma is most often clinically diagnosed as a basal cell carcinoma, verruca vulgaris, cutaneous horn, papilloma, sebaceous hyperplasia, or

squamous cell carcinoma.^{2,3} Biopsy with histologic examination of the lesion is always required for diagnosis. Histologically, desmoplastic trichilemmoma is usually a well-circumscribed lobular lesion with a central desmoplastic stroma.^{2,3} On the periphery, these neoplasms demonstrate the typical features of trichilemmoma; however, the center of the lesion is composed of irregular cords and nests of basaloid cells merged in a dense fibroblastic stroma. This often mimics an invasive carcinoma. Nevertheless, nuclear atypia and mitotic activity are usually not present. Focally, cytoplasmic pleomorphism, individual dyskeratosis, and cell necrosis may be seen.³

The extent of the invasive pseudocarcinomatous changes, as well as the overall architecture of desmoplastic trichilemmoma, makes the histopathology confused with the desmoplastic variants of basal cell carcinoma and squamous cell carcinomas. Immunohistochemical studies have shown that desmoplastic trichilemmoma epithelial cells demonstrate CD34 immunostaining, whereas this is absent in basal cell carcinomas.⁸ In the future, CD34 immunostaining may be of great value to differentiate desmoplastic trichilemmoma from basal cell carcinoma and other desmoplastic tumors as well.

Desmoplastic trichilemmoma can also be misdiagnosed as a trichilemmal carcinoma. Trichilemmal carcinoma characteristically has severe cytological atypia with mitosis and lacks the presence of stromal sclerosis.^{3,9} The reversed architectural pattern to that of desmoplastic trichilemmoma can also be found in a malignant trichilemmoma with the invasive lobules located peripherally and the central area composed of well-differentiated trichilemmal cells.³

The pathogenesis of desmoplastic trichilemmoma is not well understood. Based on histologic features and similarities with *verruca vulgaris*, some authors have believed that the trichilemmoma had an human papillomavirus-mediated etiology.^{2,3,10,11} Nevertheless, studies have not found support for a human papillomavirus-associated pathogenesis.¹²

Because desmoplastic trichilemmoma is a benign tumor, aggressive surgical excision is not required. There were no documented cases of recurrence after excision in two of the larger studies we closely examined^{2,3} or in any other studies in the literature. We found no studies that directly discussed methods of excision of the desmoplastic trichilemmoma and if Mohs micrographic surgery was utilized. Because

desmoplastic trichilemmoma is such an uncommon neoplasm rarely seen by pathologists, and because it can easily be confused with basal cell carcinoma, our pathologists recommended complete excision of the lesion. We chose to perform Mohs micrographic surgery on the patient. We believe that Mohs micrographic surgery was appropriate because of the capacity of complete margin control and because of the facial location of the lesion. There was ease of interpretation with frozen section, equivalent to that required for a basal cell carcinoma, and no special immunohistochemical stains were required. Mohs micrographic surgery allowed for maximum tissue-sparing benefits, and in our patient, Mohs micrographic surgery removal of the desmoplastic trichilemmoma had an excellent cosmetic result. Our patient remained tumor free 6 months after the procedure.

In conclusion, because these lesions often mimic invasive carcinomas and have a predilection for occurring on the face, we believe that the use of Mohs micrographic surgery is an excellent way to assure complete removal of the lesion and achieve the optimal cosmetic result.

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