Clinical Case Report

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A rare development of basal cell carcinoma on trichoepithelioma in a chemical burn scar tissue

A case report

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Abstract

Rationale: Trichoepithelioma (TE) is a rare benign skin tumor that originates from the hair follicle epithelium. Although skin lesions arising from scar tissues are mostly malignant, the development of a benign tumor such as TE is a rare event.

Patient concerns: A 28-year-old male patient who had a scar on the left cheek which arose 10 years ago because of a chemical burn visited our hospital. Scar revision was performed under local anesthesia.

Diagnosis: Histological examination identified the specimen as a TE with malignant foci suspicious of basal cell carcinoma (BCC) at the lateral margin of the specimen.

Interventions: A second stage operation was planned to excise the BCC with a 4 mm margin of the normal skin around the previous lesion.

Outcomes: No complication or recurrence was noted during the 1-year follow-up period after surgery, and the appearance of the scar improved.

Lessons: The correct differential diagnosis between TE and BCC is very important. So follow up at regular intervals is recommended for evaluation of recurrence or transformation into BCC in patients with tumors arising from chemical burn scars. And the concomitant development of TE and BCC should also be considered from a chemical burn scar.

Abbreviations: BCC = basal cell carcinoma, TE = trichoepithelioma.

Keywords: basal cell carcinoma, chemical burn, trichoepithelioma

1. Introduction

Trichoepitheliomas (TEs) are regarded as poorly differentiated hamartomas of the hair germ follicle, located mainly on the face, nasolabial folds, forehead, upper lip, and scalp. There are 2 clinical forms: hereditary multiple and nonhereditary solitary.[1] The solitary types do not show hereditary transition. Hereditary TE is seen in younger people with multiple millimetrical nodules.[2]

TE is a benign tumor but occasionally can undergo transformation to malignant neoplasms more commonly as basal cell carcinoma (BCC). Malignant transformation of such lesions is quite rare. The correct differential diagnosis between these tumors is very important because BCC is a locally aggressive neoplasm and requires total surgical excision with wide margins while TE only needs simple excision. In this article, we report a case of BCC on TE which developed after chemical burn.

2. Case report

A 28-year-old male patient visited our hospital for an examination of a scar on the left cheek which arose 10 years ago because of a chemical burn that had enlarged during the last 5 months. The scar was a 12-mm diagonally lined hypertrophic scar (Fig. 1). The patient did not report any pain or pruritus; however, he experienced slight tenderness on palpation. Scar revision was performed under local anesthesia for aesthetic purpose. On gross examination, a mass lesion was located within the scar tissue. Histological examination identified the mass as a TE located within the scar tissue that showed the typical appearances of lobules of basaloid cells and keratosis. However, a malignant focus suspicious of BCC was noted at the lateral margin of the specimen. A second stage operation was planned to excise the BCC with a 4 mm margin of the normal skin around the previous lesion. Any suspicious lateral or deep margin was confirmed to be tumor-free by frozen section biopsies during the operation. The pathological examination of the specimen revealed a 12×3 mm sized BCC at the margin of the previous scar tissue. The tumor cells showed 10 mitotic figures per 10 high-power fields and perineural invasion (Fig. 2). No complication or recurrence was

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Figure 1. A 12 mm diagonally lined hypertrophic scar with 4 mm focal depression was noticed on the left cheek.

Figure 3. Postoperative 3 months follow-up photo of the scar lesion.

noted during the 1-year follow-up period after surgery, and the appearance of the scar improved (Fig. 3). We obtained the patient's medical records and reviewed the related literature. Informed consent to participate in the study was obtained from the patient.

3. Discussion

TEs are benign skin tumors developed from skin annexes showing differentiation toward hair follicles. The lesion is usually a round-oval shaped papule or nodule, about 2 to 8 mm in diameter. The majority of the cases are found on the forehead, nasolabial groove, upper lip, and the cervical region due to the high number of sebaceous glands in these areas.[3] It may also be found on the neck, chest, upper arm, and thigh.[4] Three clinical forms of TE have been reported; a small solitary form, a small multiple forms with autosomal dominant hereditary characteristic, and a rare giant solitary form.[5,6] The management of TE by surgical excision or curettage is usually adequate due to its benign nature.

TE is histologically characterized by a well-circumscribed dermal tumor composed of uniform basaloid cells in a cellular fibrous stroma. The histopathological differentiation of TE from BCC is sometimes difficult as both neoplasms are composed of nests of basaloid cells with follicular differentiation. TE may have epithelial structures resembling hair follicle, small keratocysts lined by stratified squamous epithelium and foci of calcification while BCC consists of basaloid tumor island with peripheral palisading, stromal retraction, increased mitotic activity, and necrosis.[7,8] In some cases, the differentiation of BCC with follicular differentiation

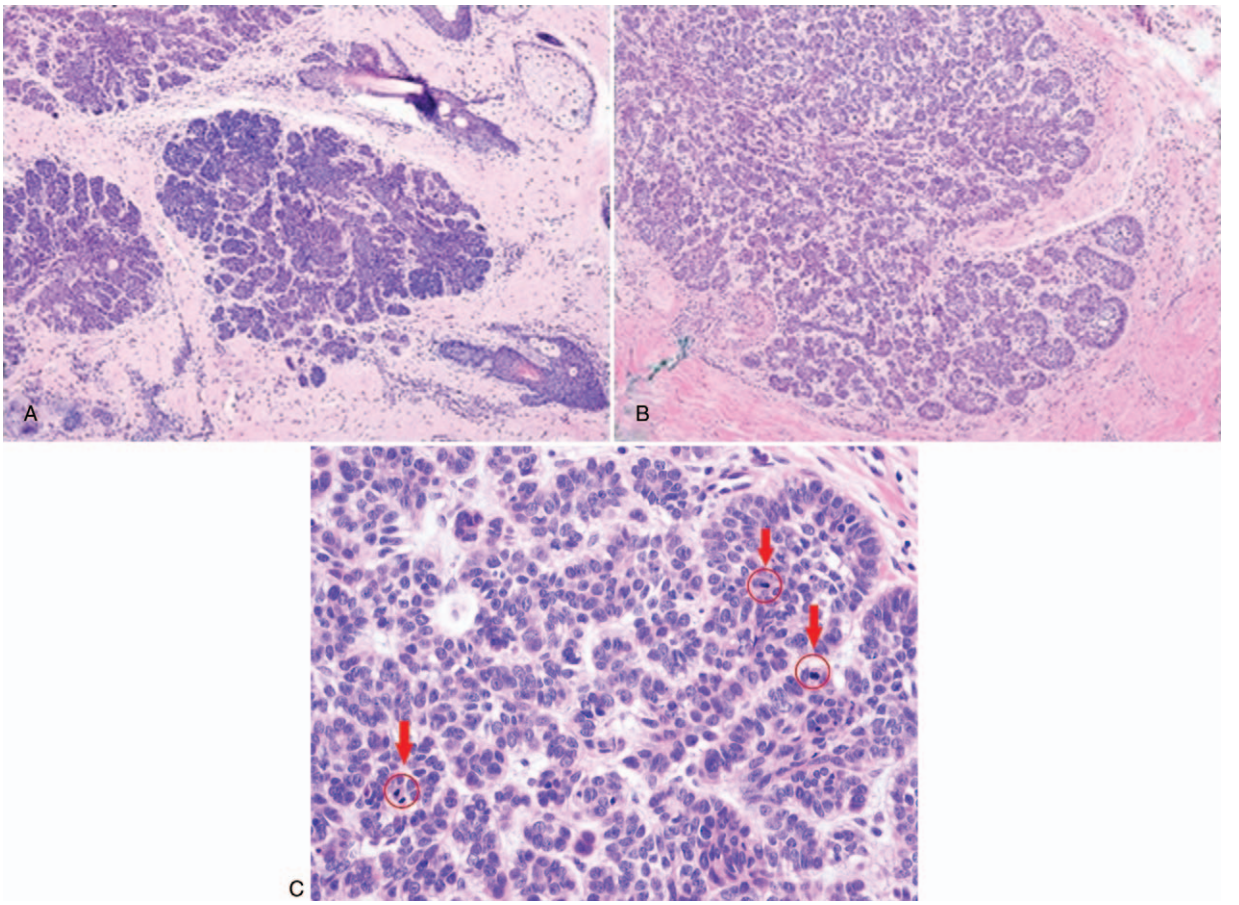

Figure 2. (A) The field shows the typical appearance of lobules of basaloid cells that form primitive hair follicle-germ structures (hematoxylin-eosin stain, ×100). Normal hair follicles are shown in the left. (B) Tumor nests shown around the nerve suspicious for perineural invasion. (C) Three mitotic figures are present (arrow).

and TE may be impossible with hematoxylin and eosin staining. Immunohistochemical staining with Bcl-2, CD10, CD34, transforming growth factor-β, Ki-67 proliferative index, pleckstrin homology-like domain, family A, member 1 (PHLDA1) may assist in distinguishing BCC from TE.[8–11]

Tumors arising from an old scar have been reported to be squamous cell carcinoma, BCC, leiomyosarcoma, malignant melanoma, clear cell adenocarcinoma, giant cell fibroblastoma, and cutaneous lymphoma.[12–14] The development of benign tumor such as TE from a scar tissue is a rare event that has been reported by Lee et al.[15] We describe a rare case of TE arising from a chemical burn scar tissue with the concomitant finding of BCC, which to our knowledge is the first to be documented. There have been reports of TE undergoing transformation to BCC usually in the setting of multiple TEs.[8,9] Sahin et al report a case of giant solitary TE which transformed into BCC.[16] However, in the case described here, the patient is a 28-year-old male with a single, small-sized TE. The age of the patient and the single lesion at presentation are not typical findings that raise suspicions for a malignant tumor. Fortunately, the patient noticed a growth in the size of the tumor in the last 5 months which can be a clinical cue to suspect a malignant lesion. Our case is unique and important in that it presents the concomitant finding of a benign and malignant tumors arising from a chemical burn scar. Follow up at regular intervals should be considered for evaluation of recurrence or transformation into BCC.[17]

4. Conclusion

Although development of a malignant tumor is a well-known complication of a chronic burn scar, benign tumor such as TE can also occur. Even in such cases, the concomitant development of BCC should also be considered. Follow up at regular intervals for evaluation of recurrence or transformation of TE to BCC is recommended in patients with tumors arising from chemical burn scars.

Author contributions

Conceptualization: Sam Yong Lee.
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Writing – original draft: Seong Hwan Kim.
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