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## Trichilemmal carcinoma in an unexposed area of the nose: A case report

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### ABSTRACT

**INTRODUCTION:** Trichilemmal carcinoma (TC) is a rare, low-grade, cutaneous malignant lesion that originates from hair follicle cells. It usually occurs in photo-exposed areas, especially on the face, scalp, neck, and dorsal part of the hand. We report a case of an adult female with TC in an unexposed area of the nose, which completely obstructed the right nasal cavity.

**CASE PRESENTATION:** An 82-year-old female presented with TC in a non-sun-exposed area of the nasal cavity, which grew progressively over one year and caused nasal obstruction. The mass had dimensions of 15cm × 8cm, and it was removed surgically with a wide and deep excision. Histopathological examination confirmed the diagnosis of TC. A follow-up at three years post-surgery revealed no signs of recurrence.

**CONCLUSION:** Surgical excision with wide margins improved the treatment outcome by preventing local recurrence and providing satisfying cosmetic results.

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### 1. Introduction

Trichilemmal carcinoma (TC) is a rare malignant tumor that is usually found in photo-exposed areas of the body, such as the scalp, forehead, neck, trunk, and upper extremities [1]. It is most commonly found in the elderly between the ages of 40 and 90 years, and the male-to-female ratio of occurrence is relatively equal [2]. According to a review by Hamman et al., only 103 cases of TC have been reported to date [3]. TC develops from the external root sheath of the hair follicle [4], and although the lesions are locally aggressive, metastases are not usually common [5]. We report a rare case of TC located in a non-sun-exposed area of the nose according to the updated consensus-based surgical case report (SCARE) guidelines

[6]. The aim of this report is to describe the diagnosis, management, and follow-up of this unusual case of TC.

### 2. Case presentation

An 82 year-old female complained of an obstructed airway due to a mass on the right nasal cavity. The mass was first noticed 5 years prior as a reddish lesion on right nasal septum. It had been growing progressively in just the past year. The patient had mild breathing difficulties due to the obstructed nose. She also struggled to engage in social activities due to the lack of confidence in her physical appearance. No weight loss was reported.

The patient's ethnicity is Minahasan (an ethnic group in Indonesia). She had a history of hypertension and is currently on amlodipine at 10 mg taken once every morning. She reported no family history of TC and no consumption of alcohol or tobacco. The patient had a biopsy one year prior, which confirmed the presence of TC. She was then scheduled for an operation to remove the tumor in the following week. However, she did not return since she lived in a rural village and had limited access to transportation.

The tumor was rooted in the nasal septum. It had dimensions of 15cm × 8cm and often bled spontaneously (Fig. 1). The tumor

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**Fig. 1.** Tumor completely blocking the right nasal cavity (blue arrow).



**Fig. 2.** A non-contrast head CT-scan showing the tumor's stalk rooted in the nasal septum (yellow arrow).

was solid in consistency, reddish in color, and well encapsulated with an uneven surface. Despite its solid consistency, any pressure applied to the tumor would cause it to bleed. Non-contrast head CT scans showed intact nasal bone and no evidence of invasion of the nasal septum (Fig. 2). No metastasis were found in the surrounding organs. Laboratory and ECG (Electrocardiography) results were within normal values.

The patient underwent elective surgery. Our team of plastic and reconstructive surgeons performed an excision and ligation of the tumor with a tumor-free margin (Fig. 3). An incision was made on the nasolabial fold until it reached the tumor's capsule. One of the challenges encountered during the surgery was carefully excising the mass while maintaining hemostasis around the capsule, as well avoiding intraoperative pulmonary aspiration from excessive bleeding in the nasal cavity.

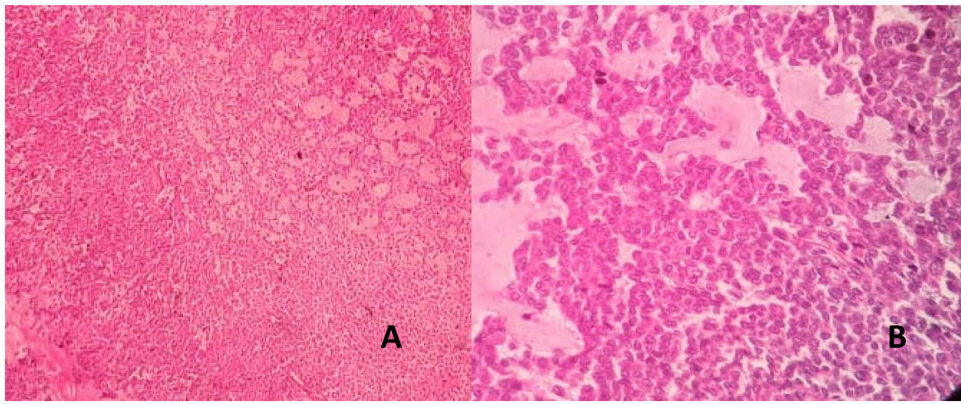
A tissue sample of the tumor was taken for biopsy. Intra-nasal mucosa was intact. The tumor branched from the medial nasal mucosa. After adequate hemostasis, the surgical area was closed using Surgicell. A post-operative tampon was placed in the nasal cavity for 3 days. An intravenous broad-spectrum antibiotic was administered for 5 days, and the patient was discharged on the 5th postoperative day.



**Fig. 3.** Intraoperative image post-tumor removal.

The tissue sample was sent to a pathology lab for examination. Hematoxylin and eosin (H & E) was used as a histopathological marker. The sample was observed under a microscope at 100x and 400x magnification. There were groups of small, diffused, round-shaped epithelial cells with large nuclei and rough chromatin bordered by hyalin connective tissues in the sample (Fig. 4). A few of the epithelial cells contained eosinophil cytoplasm and mucin. The surface contained necrotic tissues with many inflammatory cells, such as polymorphonuclear (PMN) leukocytes. The result of this second biopsy re-confirmed the diagnosis of TC.

A follow-up after 2 weeks showed prompt healing of the surgical wound, and there was no complaint of post-operative nasal



**Fig. 4.** Histopathological findings revealing TC characteristics: (A) Small groups of round-shaped epithelial cells (HE staining, magnification 4x) and (B) Prominent nuclei in epithelial cells with pale, eosinophilic cytoplasm (HE staining, magnification 40x).



**Fig. 5.** The patient tumor-free at 2 weeks post-surgery.



**Fig. 6.** The patient at 3 years post-surgery; no local recurrence of the tumor was visible.

bleeding (Fig. 5). The 3-year follow-up indicated that a satisfying cosmetic result was obtained with no evidence of the tumor's recurrence (Fig. 6). To establish an early multidisciplinary approach from the beginning in managing this patient, collaborations were established with individuals working in fields related to this case, such as clinical pathology, internal medicine, and radiology.

### 3. Discussion

TC is a rare malignancy that derives from the outer root sheath of hair follicles and usually occurs on photo-exposed, hair-bearing areas of aged individuals with fair skin [7,8]. Although multiple local recurrence is commonly found, it rarely metastasizes [9]. Its exact pathogenesis is still unknown, although it is often seen in immunocompromised patients [10]. Risk factors such as sunlight exposure play a significant role in its occurrence [9].

In previously reported cases, most TC patients had a history of long-term sun exposure [11]. In our case, the tumor was found in the right nasal cavity, which is not directly exposed to sunlight. One report found the occurrence of multiple TCs on the chest in a tuberculosis patient who underwent several partial pneumonectomies and received over 50 chest X-rays over a 30-year period, which suggests that X-ray exposure may be the trigger for non-sun-exposed TCs [12]. Garrett et al. reported another case of TC on the chest of a 59-year-old who underwent a renal transplant five years prior to developing TC. Such patients have a high risk of developing skin cancers [10]. A study in South Africa reported a 46-year-old male patient who developed recurrent TCs during treatment for breast cancer [13]. Thus, these findings indicate that prolonged sunlight exposure may not be the only risk factor for developing TC.

The diagnosis of TC is made by observing its clinical appearance and histopathologic examination [1,4]. Lesions are usually slightly raised, reddish, and ulcerated or crusted. Their clinical appearance resembles basal cell carcinoma, squamous cell carcinoma, and nodular malignant melanoma [14]. The tumor is mostly found on hair-bearing areas of the skin, and it is unlikely to grow in non-hair-bearing areas [15]. To the best of our knowledge, this is the first report of such a case. The tumor's stalk, which is the feeding artery of the tumor, arises from the inner mucosa of the right nasal septum.

Histologically, trichilemmal carcinoma is characterized by many features resembling the outer root sheath of hair follicles. Optical microscopy features include epithelial lobule proliferation centered in a pilosebaceous unit, and the lobules contain glycogen-rich bright cells [16]. Other histological characteristics of TC include prominent nuclei, translucent cytoplasm, and atypical mitotic form [1,16,17].

TC is treated by surgical excision with wide margins [16,18]. It is imperative to perform a wide excision and create a tumor-free margin to prevent local recurrence [1,9]. A study in China on surgical excisions of TCs reported that out of 26 patients, only 2 patients had local recurrence [1]. Excision and ligation of the root of the tumor located on the medial mucosa of the right nasal cavity were successfully performed in our patient. No recurrence has been reported after a 3-year follow-up. Considering that metastasis and local recurrence are rare for this type of lesion, no alternative or adjunctive therapies were applied.

### 4. Conclusion

Sunlight exposure plays an important role in the pathogenesis of rare cutaneous TC. Nevertheless, the possibility of the tumor growing in non-sun-exposed areas of the skin cannot be ruled out. Diagnosis is established through physical and histopathology examinations. Surgical excision with wide margins helps to prevent local recurrence and provides satisfying cosmetic results.

### Conflicts of interest

Nothing to declare.

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### Ethical approval

The study is exempt from ethical approval in our institution.

### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

### Author contribution

**Mendy Hatibie Oley, Deante Michelle R. Aling, and Dicky Panduwinata:** study concept and surgical therapy for this patient. **Mendy Hatibie Oley, Lily Lucia Loho, and Dicky Panduwinata:** Data collection and Writing-Original draft preparation. **Maximilian Christian Oley:** senior author and the manuscript reviewer. **Deante Michelle R. Aling, and Muhammad Faruk:** Editing and Writing. **All authors** read and approved the final manuscript.

### Registration of research studies

Not applicable – single case report.

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