Case report of a solitary fibrofolliculoma on the alar rim

Ho Yoon Jeong¹, Yong Chan Bae¹,²
¹Department of Plastic and Reconstrucive Surgery, Pusan National University School of Medicine, Busan; ²Biomedical Research Institute, Pusan National University Hospital, Busan, Korea

INTRODUCTION

Fibrofolliculoma is a benign tumor characterized by a smooth, dome-shaped papule of size 2–4 mm. Most fibrofolliculomas occur as multiple lesions, and very rarely, they are solitary. Herein, we report a case of solitary fibrofolliculoma found in the alar rim, without the typical characteristics of a fibrofolliculoma. A 42-year-old man visited the hospital with a protruding lesion that had occurred 1 year previously. A mass of size 5×7 mm was observed on the left alar rim. The tumor was dome-shaped and palpable. The patient did not have any similar lesions elsewhere. No family member was known to have such a lesion. An incisional biopsy was performed before surgery, and pathological examination revealed hyperkeratosis and dyskeratosis; however, an accurate diagnosis was not made. Complete resection was planned for the mass on the alar rim. The resected mass was subjected to permanent biopsy, and the pathological examination results led to the diagnosis of fibrofolliculoma. Therefore, when diagnosing a dome-shaped mass in the alar rim, despite the suspicion of a very rare disease, it is necessary to suspect fibrofolliculoma and consider the process from diagnostic examination to treatment.

Keywords: Hair diseases / Nose / Skin neoplasm

Case Report

A 42-year-old man presented to the hospital with a palpable mass, which had appeared a year ago, on his left alar rim (Fig. 1). The mass was 5 × 7 mm in size, protruding and palpable, dome shaped, and the same color as that of his skin. He did not have...
similar lesions in other areas of his body. In addition, none of his family members were known to have such a lesion. The patient had visited a local hospital before consulting in our clinic and had been diagnosed with keratoacanthoma. Under this diagnosis, incisional biopsy was performed on a portion of the mass for histological confirmation, along with the removal of the lesion. Pathologic examination revealed hyperkeratosis and parakeratosis, and complete excision was planned. Under local anesthesia, an elliptical incision was made to remove the lesion, including the mass. When viewed with the naked eye, no invasion of the mass into cartilage or other tissues was observed, and no adhesion to the periphery was observed. Therefore, the mass was considered to have been completely excised above the perichondrium. Permanent biopsy was requested for the excised mass, and nevus sebaceous was confirmed. We requested the pathology department for a review because it did not seem clinically correct, as nevus sebaceous is a congenital malformation that can be found on the face [12]. Histological examination revealed the presence of proliferative follicular epithelium surrounded by perifollicular fibrous tissue (Fig. 2). Thin epidermal strands originated from the hair follicles and centered around the infundibulum of the hair. The connective tissue around the epithelium also showed sclerotic features (Fig. 3). Based on these pathological results, the final diagnosis was fibrofolliculoma.

**DISCUSSION**

Several tumors are formed in hair follicles [13-16]. Clinically, it is very difficult to distinguish fibrofolliculoma from trichodiscoma and perifollicular fibroma. When viewed with the naked

---

**Table 1.** Previously reported cases of solitary fibrofolliculoma

| Author               | Case | Age (yr)/sex | Location     | Tumor size            | Mass appearance                      |
|----------------------|------|--------------|--------------|-----------------------|--------------------------------------|
| Scully et al. [3]    | 1    | 65/F         | Chin         | 5 mm in diameter      | Skin-to-pink colored, dome-shaped papule |
| Starink and Brownstein [5] | 2    | 49/M         | Chin         | 5 mm in diameter      | Yellowish nodule                      |
|                      | 3    | 20/F         | Nose         | Skin-colored papule   |                                      |
|                      | 4    | 50/M         | Cheek        | Skin-colored papule   |                                      |
|                      | 5    | 60/F         | Ear          | 3 mm in diameter      | Dome-shaped papule                   |
|                      | 6    | 52/M         | Eyebrow      | 6 × 4 mm              |                                      |
| Hong et al. [8]      | 7    | 40/F         | Scalp        | 7 × 6 × 5 mm          | Skin-to-pink-colored papule          |
| Chang et al. [2]     | 8    | 37/F         | Eyelid       | 5 × 5 mm              | Skin-colored papule                  |
| Cesinaro et al. [6]  | 9    | 63/F         | Nose         | 12 × 10 mm            | Skin-colored papule                  |
| Cho et al. [9]       | 10   | 45/M         | Ear          | 12 × 10 × 8 mm        | Flesh-colored mass                   |
| Criscito et al. [10] | 11   | 72/F         | Cheek        | 4 mm in diameter      | Flesh-colored, dome-shaped papule    |
| Sohn et al. [11]     | 12   | 50/M         | Posterior auricular region | 10 × 12 mm | Flesh-colored papule                  |
| Wang and Cheng [1]   | 13   | 68/F         | Eyelid       | 5 × 5 × 4 mm          | Flesh-colored papule                  |
| Mishra et al. [4]    | 14   | 50/M         | Eyelid       | 3 × 3 × 2 cm          |                                      |

M, male; F, female.

**Fig. 1.** A 42-year-old man with a 5×7 mm mass on the left alar rim.

**Fig. 2.** Microscopic image of the lesion showing proliferative follicular epithelium (black arrows), surrounded by perifollicular fibrous tissue (black arrowheads) (H&E, ×12.5).
eye, all these lesions are pale-yellow or white, 2–4 mm in size, usually dome shaped, and smooth [3]. Furthermore, according to the cases of solitary fibrofolliculoma reported thus far, most of them occurred in patients aged 50 years or older, and unlike multiple fibrofolliculomas, they do not show a genetic tendency and do not accompany other skin diseases [5]. To date, the only method for obtaining a definitive diagnosis of fibrofolliculoma is pathology. Therefore, diagnosis is made according to the pathological examination of an incisional biopsy and clinical judgment at presentation. In addition, solitary fibrofolliculoma is very rare; hence, there are not many clinical cases, and because it is difficult to distinguish between clinically similar types of perifollicular fibromas and trichodiscomas, it is highly likely to be misdiagnosed. Therefore, biopsy should be performed. Histologically, fibrofolliculoma is represented as a circular or oval-shaped well-bounded fibroepithelial proliferative tumor in the dermis, which appears as a concentric circle around the hair follicle. In the center of the tumor, epithelial proliferation of a single follicular infundibulum is generally observed and forms an epithelial zone that extends into the surrounding fibrous matrix, leading to unique network formation by anastomosis [8,17]. In this case, the lesion had thin epidermal strands centered around the infundibulum of the hair. In the center of the tumor, epithelial proliferation of a single follicular infundibulum is generally observed and forms an epithelial zone that extends into the surrounding fibrous matrix, leading to unique network formation by anastomosis [8,17]. In this case, the lesion had thin epidermal strands centered around the infundibulum of the hair and the proliferative follicular epithelium, surrounded by connective tissue. In the case of multiple fibrofolliculomas, laser therapy using Erbium-YAG or CO₂ and systemic isotretinoin administration are reportedly effective; nevertheless, treatment results vary [18]. Because of their rarity, there is no consensus on the treatment and prognosis of solitary fibrofolliculoma. Even though solitary fibrofolliculoma is known to be a benign mass, complete resection is considered desirable. In addition, as the protruding mass changes the shape of the alar rim, it was judged that complete resection is more desirable than other treatment methods to provide the patient a contouring effect for esthetic purposes. We report the diagnosis and treatment of a solitary fibrofolliculoma, a rare disease, in a healthy male with no specific medical history or family history of mass development in the nose. Fibrofolliculoma is a rare disease; however, when a mass occurs in the nose, clinicians should suspect fibrofolliculoma based on the clinical and histological characteristics presented in this case and consider treatment based on diagnostic tests and complete resection.

NOTES
Conflict of interest
Yong Chan Bae is an editorial board member of the journal but was not involved in the peer reviewer selection, evaluation, or decision process of this article. No other potential conflicts of interest relevant to this article were reported.

Ethical approval
The study was approved by the Institutional Review Board of Pusan National University Hospital (IRB No. 2106-018-104) and performed in accordance with the principles of the Declaration of Helsinki. Written informed consent was obtained.

Patient consent
The patient provided written informed consent for the publication and the use of his images.

ORCID
Ho Yoon Jeong https://orcid.org/0000-0003-1837-9146
Yong Chan Bae https://orcid.org/0000-0002-0268-4667

Author contribution
Conceptualization: YCB. Writing - original draft: HYJ. Writing - review & editing: YCB, HYJ. Supervision: YCB.

REFERENCES
1. Wang W, Cheng J. Solitary fibrofolliculoma of the upper eyelid in a 68-year-old female: a case report. BMC Ophthalmol 2020; 20:97.
2. Chang JK, Lee DC, Chang MH. A solitary fibrofolliculoma in the eyelid. Korean J Ophthalmol 2007;21:169-71.
3. Scully K, Bargman H, Assaad D. Solitary fibrofolliculoma. J Am Acad Dermatol 1984;11(2 Pt 2):361-3.
4. Mishra DK, Jakati S, Kaliki S. Solitary fibrofolliculoma of the eyelid: a less known entity. Indian J Pathol Microbiol 2021;64: 195-6.
5. Starink TM, Brownstein MH. Fibrofolliculoma: solitary and multiple types. J Am Acad Dermatol 1987;17:493-6.
6. Cesinaro AM, Rusev BC, Kutzner H. Fibrofolliculoma with ancient/pseudosarcomatous features. J Cutan Pathol 2010;37: 987-90.
7. Weintraub R, Pinkus H. Multiple fibrofolliculomas (Birt-Hogg-Dubé) associated with a large connective tissue nevus. J Cutan Pathol 1977;4:289-99.
8. Hong JK, Yoon DH, Kim TY, Kim HO, Kim CW. A case of solitary fibrofolliculoma. Ann Dermatol 1997;9:286-8.
9. Cho E, Lee JD, Cho SH. A solitary fibrofolliculoma on the concha of the ear. Int J Dermatol 2012;51:616-7.
10. Criscito MC, Mu EW, Meehan SA, Polsky D, Kopeloff I. Dermoscopic features of a solitary fibrofolliculoma on the left cheek. J Am Acad Dermatol 2017;76(2S1):S8-9.
11. Sohn KM, Woo YJ, Kim JE, Kang H. A solitary nodule on the posterior pinna. Indian J Dermatol Venereol Leprol 2019;85: 111-3.
12. Idriss MH, Elston DM. Secondary neoplasms associated with nevus sebaceus of Jadassohn: a study of 707 cases. J Am Acad Dermatol 2014;70:332-7.
13. Wee SJ, Park MC, Chung CM. Basal cell carcinoma misdiagnosed as trichoepithelioma. Arch Craniofac Surg 2020;21:202-5.
14. Hu JL, Yoo H, Kwon ST, Kim S, Chung JH, Kim H, et al. Clinical analysis and review of literature on pilomatrixoma in pediatric patients. Arch Craniofac Surg 2020;21:288-93.
15. Choi JY, Cha WJ, Kwon HJ, Seo BF, Kwon H, Jung SN. A giant solitary vellus hair cyst on the nasal root. Arch Craniofac Surg 2020;21:326-8.
16. Koh IS, Cho HJ, Kim JW. Rapidly growing giant pilomatrixoma in the right parotid region of a pregnant woman. Arch Craniofac Surg 2020;21:176-9.
17. Birt AR, Hogg GR, Dube WJ. Hereditary multiple fibrofolliculomas with trichodiscomas and acrochordons. Arch Dermatol 1977;113:1674-7.
18. Welsch MJ, Krunic A, Medenica MM. Birt-Hogg-Dubé syndrome. Int J Dermatol 2005;44:668-73.