皮脂腺髮毛囊瘤

—病例報告—

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Sebaceous Trichofolliculoma

—A Case Report—

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Sebaceous trichofolliculoma is rare. We herein report a case of sebaceous trichofolliculoma in an 18-year-old boy. The tumor presented as an asymtomatic , elastic, dermal nodule with a central opening at the nasal bridge near the left inner canthus. Histologically, the tumor showed a wide branching cavity lined by keratinizing epithelium connected to the surface. There were keratin, corneocytes and hair shafts in the dilated cavity. Radial to the central cavity were abundant well-differentiated sebaceous lobules, sebaceous ducts and hair follicles in various stages of the hair growth cycle. (Dermatol Sinica 19：357-361, 2001)

Key words: Sebaceous trichofolliculoma, Trichofolliculoma, Folliculosebaceous cystic hamartoma

皮脂腺髮毛囊瘤爲一罕見腫瘤。我們報告一例發生在一位十八歲男性的皮脂腺髮毛囊瘤。此腫瘤位於鼻樑靠近左側內眼眥處，爲一無症狀具彈性的眞皮小結節，中心在表皮有一小開口。組織學下此腫瘤中心爲一連接到表皮的分枝狀空腔，爲角化上皮所覆蓋；空腔內有角質，角質細胞及毛髮。空腔向外輻射出許多分化良好的皮脂腺，皮脂腺管及處於各種不同生長階段的毛囊。(中華皮誌19：357-361, 2001)

INTRODUCTION

Sebaceous trichofolliculoma (STF) was first reported by Plewig in 1980 as a centrally depressed lesion on the nose with hairs emerging from sinus-like openings.[1] Histologically, abundant sebaceous lobules associated with hair-bearing units were directly connected to the epithelial lining of a dilated central cavity. In this report, we describe a case of STF.

CASE REPORT

An 18-year-old Taiwanese boy visited our

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hospital in August 1999 with the complaint of an asymptomatic slowly growing facial tumor noted for 2 years. He was otherwise healthy. Physical examination revealed a movable, elastic, dome-shaped, bean-sized, skin-colored dermal nodule with a central opening at his nasal bridge near the left inner canthus (Fig. 1). No hair emerged from the central opening. Epidermal cyst was impressed clinically. Total excision of the tumor was performed under local anesthesia.

Histologically, it revealed a circumscribed non-encapsulated dermal tumor with a dilated branching cavity lined by keratinizing squamous epithelium. The cavity was connected to the surface via a dilated pore, corresponding to the central opening clinically. There were keratinous material, corneocytes and hair shafts in the central cavity (Fig. 2). Radial to the cavity were many mature sebaceous lobules connected to the central cavity via sebaceous ducts and hair follicles in different stages of the hair growth cycle (Fig. 3, 4). Bizarre-shaped secondary and tertiary hair follicles with differentiation towards lower segments of follicles were noted (Fig. 4). There was a fibrous stroma surrounding the epithelial component of the tumor. No adipose component was present in the fibrous stroma (Fig. 3). A final diagnosis of STF was made. There was no sign of recurrence 18 months after surgery.

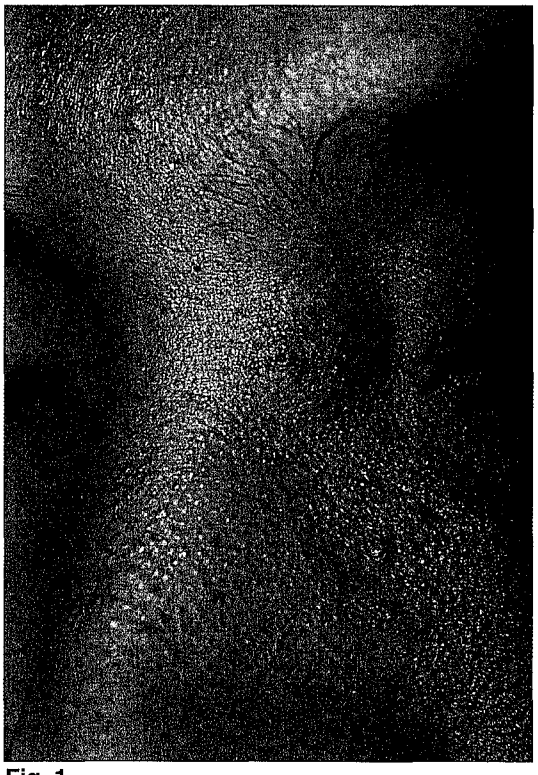

Fig. 1
There was a dome-shaped dermal nodule with a central opening at the nasal bridge near the left inner canthus.

DISCUSSION

STF is a hamartomatous adnexal tumor first reported by Plewig as a variant of trichofolliculoma (TF) in 1980.[1] There were only a few reports of STFs after Plegwig's report.[2-8] However, most of the reported cases are now considered as folliculosebaceous cystic hamartoma (FSCH),[9] a recently identified entity by Kimura et al. in 1991.[9]

TF usually locates on the face and presents as a small, skin-colored, dome-shaped nodule with a central pore.[10,11] Histologically, it consists of a dilated cyst lined by keratinizing squamous epithelium and contains keratinous material and hair shafts in the cyst.[10,11] Radiating from the cyst are many fairly differentiated secondary follicles. By definition, TF should not have sebaceous lobules. Focal sebaceous differentiation can be found in some TFs but it is

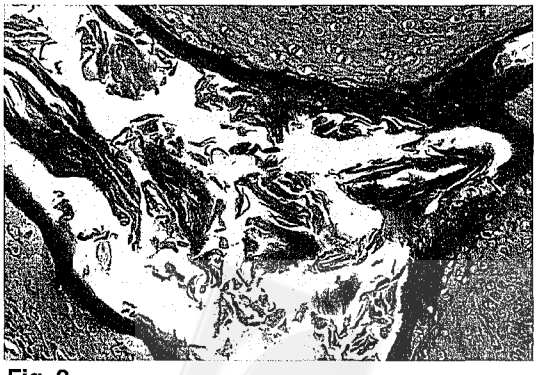

Fig. 2
There were keratin and hair shafts in the central cavity. The central cavity was lined by keratinizing squamous epithelium. (H & E, x200)

not a prominent feature.[11] The sebaceous lobules in a TF are small and never reach the size of sebaceous lobules in sebaceous follicles.[1,11]

STF shares many similar clinical and histological features with TF. STF usually locates on the face, too.[1] It presents as a centrally depressed lesion with hairs emerging from a sinus-like opening. Histologically, it is a non-encapsulated tumor located mostly in the dermis. It is similar to TF by the presence of a centrally located cystic cavity lined by keratinizing squamous epithelium and contains keratin and hair shafts in the cavity.[1] The cavity is connected

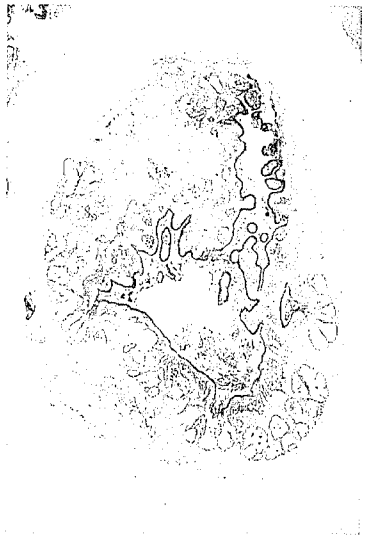

Fig. 3
Radial to the central cavity were many mature sebaceous lobules connected to the central cavity via sebaceous ducts. The surrounding fibrous stroma was devoid of adipose component. (H & E, x10)

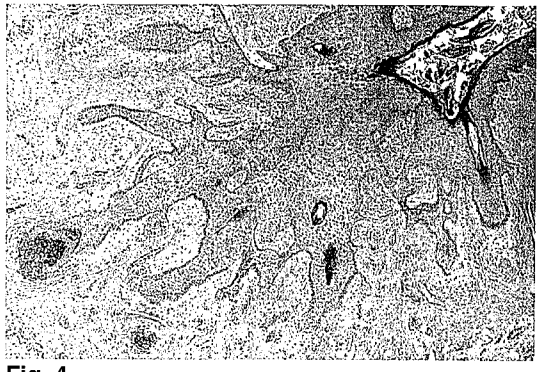

Fig. 4
There were bizarre-shaped secondary and tertiary follicles in different stages of the hair growth cycle. Differentiation towards lower segments of hair follicles and formation of hair shafts were also noted. (H & E, x40)

Table I. Summary of histological differential diagnoses of sebaceous trichofolliculoma

Differential diagnoses	Differential features
Sebaceous trichofolliculoma	Hair shafts in the central cyst Secondary follicles with differentiation towards lower segments of hair follicles Prominent sebaceous lobules
Folliculosebaceous cystic hamartoma	No hair shafts in the central cyst No differentiation towards lower segments of hair follicles Characteristic mesenchymal change
Trichofolliculoma	No prominent sebaceous lobules
Cystic sebaceous hyperplasia	No secondary hair follicles
Dermoid cyst	Eccrine and apocrine glands, smooth muscle fibers, and small nerve fibers surrounding the cyst wall

to the epidermis via a dilated ostium.[1] There are also secondary hair follicles in various stages of the hair growth cycle radial to the dilated cavity.[1] The most prominent feature of STF is the presence of abundant mature sebaceous lobules connected to the epithelium of the cavity via sebaceous ducts.[1] A fibrous stroma surrounds the epithelial component of the tumor.

FSCH, TF, sebaceous hyperplasia, and subcutaneous dermoid cyst should be differentiated from STF. The histological differential diagnoses are summarized in Table I.

FSCH is a recently defined entity by Kimura *et al.*[9] It is usually pedunculated or dome-shaped without a central dilated pore and is located on the face or scalp.[9,12,13] Histologically, it consists of an fibroepithelial component and a distinct mesenchymal change.[9,12,13] The fibroepithelial component shows an infundibular cystic structure attached to well-differentiated sebaceous lobules via sebaceous ducts and compactly laminated fibroplasias around the entire epithelial component of fibroepithelial units.[9,12,13] Most of the dilated infundibular cysts have no connection to the epidermis.[9,12,13] Its distinct mesenchymal change surrounds the fibroepithelial component and consists of fibrillary bundles of collagen, an increased number of small venules and adipocytes.[9,12,13] It can be differentiated from STF by the absence of follicular structures differentiating towards lower segments of follicles, the absence of hairs in the central cavity, and the presence of characteristic mesenchymal change.[9,12,13]

Usually, sebaceous hyperplasia can be easily identified clinically as small soft, yellow, slightly umbilicated papules on the face in persons past middle age.[14] Rarely, sebaceous hyperplasia may clinically manifest as an intracutaneous nodule with a central pore and histologically display a cystic structure with a dilated infundibulum.[15] However, it can be easily differentiated from STF by the absence of secondary hair follicles.[15]

A subcutaneous dermoid cyst is usually present at birth along lines of embryonic fusion.[16] On the face, it is usually located at the lateral aspect of the upper eyelid. It is seated deeply in the subcutaneous tissue and is often adherent to the periostium. Though there are follicles and sebaceous glands in the wall and hair shafts in the lumen, it can be differentiated from STF by the presence of eccrine glands, apocrine glands, smooth muscle fibers, small nerve fibers and vascular stroma surrounding the cyst.[16]

Whether STF is a variant of TF is of debate.[1,17] Some authors consider STF as a variant of FSCH.[18,19] STF, FSCH and TF are even deemed as different spectrum of the same pathological process by Schultz and Hartschuh.[20] They divide TFs into 3 stages according to the evolutional changes: fully developed TF, TF at late stage and TF at a very late stage.[20] In their model of the evolution of TFs, the inferior follicular elements of follicles undergo regressive changes while development and maturation of sebaceous elements occur.[20] Adipocytes can often be shown in the stroma at the late stage of TF, indicating a reduction of follicular epithelium and perifollicular sheath and a sign of aging of the lesion.[20] Hence, they consider STF as a TF at a late stage and FSCH as a TF at a very late stage.[20] However, chronological changes of the above tumors are difficult to substantiate because these tumors are usually excised completely and FSCH and STF are much rarer as compared with TF. These appendage tumors frequently present with overlapping features. However, if the salient features of each tumor can be better appreciated , most tumors can be differentiated histologically.

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