

Multiple Trichoepithelioma Associated with Milia

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We present a case of multiple trichoepithelioma associated with milia. A 12-year-old girl developed symmetrically distributed, numerous, skin-colored papules on her face, some of which were studded with milia-like whitish materials. Histopathological findings of these lesions revealed the typical findings of trichoepitheliomas with overlying milia in the upper dermis. (*Ann Dermatol* 10:(3) 172~174, 1998).

Key Words : Multiple trichoepithelioma, Milia

Milia-like eruptions have been described as a sign of syringoma^{1,2}, cutaneous calculi³ and pilomatricoma⁴. Among them two cases of syringomas also displayed histologically milia-like large cysts filled with keratin located directly beneath the epidermis^{1,2}. However, despite the usual presence of keratin-filled cysts histologically in trichoepitheliomas, there has been no case report that showed, clinically and histologically, the overlapping features of trichoepitheliomas and milia in the same skin lesion. We describe a patient with non-hereditary multiple trichoepitheliomas on the face, some of which were studded with milia-like whitish materials. A histopathological examination of these lesions revealed the typical findings of trichoepitheliomas with overlying milia in the upper dermis. The literature about the relationship between multiple trichoepitheliomas and milia are reviewed.

CASE REPORT

A 12-year-old girl presented with a 6-year history of symmetrically distributed, multiple, asymptomatic, round papules located on the face, especially on the nose, nasolabial folds, cheeks and upper eyelids. The lesions were flesh-colored and had a

firm consistency. Their size varied between 2 mm and 10 mm in diameter. Several lesions had whitish material like kernels of rice on the surface, clinically suggesting milia (Fig. 1). A physical examination was otherwise normal. The patient had no family history of these types of the lesions.

Multiple punch biopsies were taken from various lesions. A histopathological examination revealed dermal tumors commonly composed of islands of basophilic cells showing peripheral palisading, arranged as solid aggregates, and multiple horn cysts with a fully keratinized center surrounded by basophilic cells (Fig. 2). The lesions with whitish materials revealed milium-like large cysts in the upper dermis (Fig. 3). These cystic structures were lined by a stratified epithelium of a few cell layers and contained concentric lamellae of keratin. There were some connections between the upper milium-like large cyst and the lower tumor islands. However, no specimen revealed milium-like larger cysts without tumor islands.

She was treated with electro-surgery three times, but three months later recurrence was noted in some of the lesions.

DISCUSSION

Multiple trichoepitheliomas may be familial with an autosomal dominant mode of inheritance⁵. Clinically, they usually appear as numerous, small, flesh-colored papules usually between 2 and 8 mm and located mainly along the nasolabial folds but also on the forehead, nose, and upper lip.

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Fig. 1. Symmetrically distributed, multiple, flesh colored papules and nodules located on the nose, nasolabial folds, cheeks and upper eyelids, some of which were studded with milia-like whitish materials (arrow).

Fig. 3. A histopathological examination taken from the lesions with whitish materials revealed the overlying milium-like larger cysts associated with trichoepithelioma in the upper dermis (H & E, $\times 40$).

Histopathological findings consist of tumor islands of basophilic cells with peripheral palisading and multiple horn cysts. Although this patient had no family history of similar lesions, the clinical and histopathological findings were well consistent with multiple trichoepithelioma. However, this case appears to be unique, in that some of the trichoepithelioma lesions were studded with whitish material like kernels of rice and a few lesions in the areas containing vellus hairs such as the eyelids and cheeks looked like milia rather than trichoepitheliomas. Microscopic examination of these lesions showed large keratin-filled cysts in the upper

Fig. 2. A histopathological examination revealed the typical findings of trichoepitheliomas composed of islands of basophilic cells showing peripheral palisading, arranged as solid aggregates, and multiple horn cysts with a fully keratinized center surrounded by basophilic cells (H & E, $\times 40$).

dermis overlying the trichoepitheliomatous lesions.

Two interpretations of the relationship between trichoepitheliomas and milia in our case are possible. One possibility is that the trichoepitheliomas and milia developed independently and some of them were coincidentally seen in the same lesion. This explanation seems unlikely as we found no specimen showing milia alone without trichoepitheliomatous components, although multiple biopsies were taken from various lesions. The other possible interpretation is that milia developed in association with or by transformation from trichoepitheliomatous components. The histopathological finding that a part of milium was connected by a pedicle of basal cells to the tumor islands of trichoepitheliomas supports this possibility. Hence, we formulated the diagnosis of multiple trichoepitheliomas associated with milia.

The coexistence of trichoepitheliomas and milia in a patient have been observed occasionally in the literature. There have been some reports on a syndrome of the simultaneous appearance of multiple trichoepitheliomas, milia and cylindromas in a patient with a family history^{6,7}. In addition, the association of milia and epidermal cysts in a patient with desmoplastic trichoepithelioma was described⁸. D'Souza et al⁹. also reported a case of multiple trichoepithelioma in association with epidermal cysts.

Histologically, the trichoepitheliomas and milia share common features, including cystic structures

composed of peripheral basaloid cells, a thin granular layer, laminated and keratinized centers, fronds of basaloid cells, and fibroblastic stroma⁶. They differ in the proportion of keratinized to basaloid cells and in the size of each lesion. In the milia, the keratinized component is predominant, and this may account for its yellow-white appearance clinically. Based on this histological resemblance, Thies and Schwarz¹⁰ speculated that milia seem to be miniature trichoepitheliomas that have been modified in the direction of vellus hair formation. Recently, van der Putte⁷ examined 250 serial specimens from 14 members of a family with inherited multiple trichoepitheliomas, milia, cylindromas, and spiradenomas. In some specimens of the trichoepitheliomatous lesions, increasingly larger variants of keratin cysts formed a series ending in milia, which demonstrated their derivation from a trichoepitheliomatous bulge proliferation. However, excluding our patient, there has been no mention of distinctive clinical and histological evidence of overlapping features of trichoepithelioma and milia in the same lesion. This case and others suggest that there might be a close relationship between milia and trichoepitheliomas in their histogenesis.

REFERENCES

1. Friedman SJ, Butler DF: Syringoma presenting as milia. *J Am Acad Dermatol* 16:310-314, 1987.
2. Weiss F, Paez E, Greenberg AS, et al: Eruptive syringomas associated with milia. *Int J Dermatol* 34:193-195, 1995.
3. Neild VS, Marsden RA: Pseudomilia: widespread cutaneous calculi. *Clin Exp Dermatol* 10:398-401, 1985.
4. Lucky AW, Prendiville JS: Centrofacial milia-like eruption. *Pediatric Dermatology* 10:382-4, 1993.
5. Elder D, Elenitas R, Ragsdale BD: Tumors of the epidermal appendages. In Elder D, Elenitas R, Jaworsky C (eds): *Histopathology of the skin*. 8th ed. Lippincott-Raven, Philadelphia, 1997, pp747-803.
6. Rasmussen JE: A syndrome of trichoepitheliomas, milia and cylindromas. *Arch Dermatol* 111:610-614, 1975.
7. van der Putte SC: The pathogenesis of familial multiple cylindromas, trichoepitheliomas, milia, and spiradenomas. *Am J Dermatopathol* 17:271-280, 1995.
8. Sumithra S, Jayaraman M, Yesudian P: Desmoplastic trichoepithelioma and multiple epidermal cysts. *Int J Dermatol* 32:747-748, 1993.
9. D'Souza M, Garg BR, Ratnakar C, et al: Multiple trichoepitheliomas with rare features. *J Dermatol* 21:582-585, 1994.
10. Thies W, Schwarz E: Multiple eruptive millien-ein organoides follikhamartom. *Archiv Klin Exp Dermatol* 214:21-34, 1961.

1. Friedman SJ, Butler DF: Syringoma presenting as