Milia En Plaque
- A Case Report
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‘Milia en plaque’ is a rare disease entity with unclear pathogenesis but distinctive histopathology. No more than 30 cases have been recorded in the world’s literature. Most of them occurred in the peri-auricular area. There were no optimal treatments. We describe a 65-year-old man with a flesh-colored plaque embedded with tiny globid cysts on his left cheek, which developed in one year. Combined its clinical and histopathological findings, we concluded it as milia en plaque and treated it with 3-month oral minocycline. Almost complete regression was gained. This experience again provided the information that oral minocycline appears to be an effective and inexpensive method in treating the milia en plaque. (Dermatol Sinica 26: 180-184, 2008)

Key words: Milia en plaque, Treatment, Oral minocycline

INTRODUCTION
Milia are rather common skin tumors and are classified as primary or secondary. Primary milia are derived from the vellus hair and the secondary milia present represent retention cysts derived from hair follicles and eccrine ducts.¹ Subepidermal bullous dermatoses, dermabrasion, ionizing radiation and topical agents such as steroid or 5-fluorouracil treatment were proposed to be the triggering factors of secondary milia.¹ Milia en plaque is a quite rare variant of milia, which occurs spontaneously without identifiable causative factors. In this article, we reviewed its differential diagnoses, postulated pathogenesis and managements.

CASE REPORT
A 65 years old man presented with the complaint of a facial indurated plaque without marked discomfort for 1 month. The lesion first appeared as an itching, mild swelling and erythematous plaque on his left cheek. He denied progressive body weight loss, fatigue, fever, joint discomfort, photosensitivity, topical medication, antecedent vesiculobullous disease and ionizing exposure. There was no contributory family history. He was treated as chronic photodermatitis with potent topical corticosteroid (mometasone furoate ointment, 0.1%) in the first 5 months. The treatment response was poor. He was lost to follow up for the next 6 months and in June, 2007, he came back to our clinic and the lesion’s size remained the same but became hyperpigmented. He received an incisional biopsy.

On physical examination, there were...
no periungual telangiectasia, oral ulcer, swollen joint or permanent hair loss. The cutaneous findings revealed a pigeon-egg sized erythematous and hyperpigmented indurated plaque studded with tiny yellowish papules on the left cheek (Fig. 1A, 1B) Telangiectasia was also noticed. The laboratory data including CBC, ESR, BUN, creatinine, urine routine examination and ANA titer were all within the normal ranges. Biopsy from left cheek was taken and the pathology demonstrated several small keratin-filled cysts just beneath the epidermis. These cysts were enveloped by the stratified squamous epithelium and surrounded by a moderate lymphocyte infiltrate (Fig. 2) We treated the patient with oral minocycline, 100mg twice a day and topical adapalene gel (0.01%). Adapalene was discontinued one month later due to local irritancy and the lesion resolved in 3 months. (Fig. 3)

DISCUSSION

Balzer and Fouquet first described the clinical manifestations of plaques with confluent tiny cysts on bilateral post-auricular area in 1903. The condition was named by Hubler et al. in 1978. The clinical picture is characterized by tiny yellowish globoid cysts embedded in an erythematous base.

The pathogenesis of milia en plaque remains unclear. Several trigger factors include glasses, perfume and even the cold environment were proposed. However, the mechanism remained unestablished in the majority of cases. Ishiura et al. discovered that the lesions stained positively with cytokeratin 6, 16 and expressed increased proliferating cell nuclear antigen. This may explain a part of pathogenesis of milia en plaque.

Despite the majority of cases of milia en plaque are of unknown pathogenesis, milia en plaque associated with underlying diseases such as discoid lupus erythematosus and pseudoxanthoma elasticum have been reported. Some authors observed that milia en plaque were coexistent with mycosis fungoides and follicular mucinosis. All of these reported cases revealed some degree of antecedent tissue injury by the underlying diseases and attributing to the following development of milia en plaque. Boehm et al. proposed damage to the adnexal structures may induce milia formation and the follicular plugging found in discoid lupus erythematosus might be a cause. Kouba et al. believed milium formation in their case was most likely to be attributable to alteration of the
infundibular portion of the follicle, resulting in dysfunctional keratinization and outlet obstruction. Cho et al. considered that the degeneration of elastic fibers result in the formation of milia en plaque. Another report described the eruption on the leg in the renal transplant patient who received 2 years of cyclosporine and considered it as adverse effect of cyclosporine. Consequently, they were then categorized to the “secondary milia en plaque” entity in order to differentiate from those without definite causative factors.

Several dermatoses resemble similar clinical manifestations of milia en plaque. The differential diagnoses of the clinical pattern include lichen planus tumidus follicularis described by Belaich et al. in 1977. Histologically, it was characterized with the picture of follicular lichen planus in which follicles and cysts surrounded by lichenoid infiltration. The other considered entity was steroid rosacea which was characterized by vascular dilatation of upper and mid-dermal vessels with perivascular and perifollicular lymphohistiocytic inflammation. Besides, rosacea contains no comedones. Favre-Racouchot syndrome, localized steatocystoma multiplex were excluded by the clinical manifestations and histopathology.

Most of the cases occurred on peri-auricular area. Other reported sites included eyelids, ear lobes, periorbital area, cheek, submandibular area, supraclavicular area, mental area and even the lower leg. The mean age of affected patients was 44 years (range from 33-65 years) and there was a female predilection (female/male=3/1). No familial inheritance was noticed.

There is no standard treatment for milia en plaque, but it can be treated by either surgical or medical modalities. The reported effective surgical modalities included simple evacuation with incision, electrodessication, dermabrasion and CO2 laser evaporation. However, scarring or pigmentary changes can result from these surgical approaches. Medical treatment of milia en plaque, including topical tretinoin, oral isotretinoin, oral etretinate, oral minocycline, or photodynamic therapies, produces variable responses. Spontaneous regression has also been documented. In general, topical tretinoin or extraction was effective in superficially located milia, while minocycline worked in cases with dense inflammatory infiltrates in dermis.

![Fig. 2](image)

Cysts were lined by stratified squamous epithelium and filled with laminated keratin in the dermis. (H&E, original magnification x40)

![Fig. 3](image)

Marked reduction in telangiectasia and the number of milia.
In conclusion, milia en plaque is a rare disease without known optimal treatments. Oral minocycline, 100mg twice daily for 3 months achieved good outcome with almost complete resolution of our case and proved the usefulness of this non-invasive modality in cases even without marked cellular infiltrate.

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粟粒疹板塊
-病例報告

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粟粒疹板塊為一極少見之疾病，雖然致病機轉目前尚未釐清，但卻有其獨特之病理特徵。至今未超過30例被報導於世界文獻之中。大部分的病灶都發生在耳朵週遭。目前此病仍未有最佳的處理方式。我們於此報告一位65歲患者於1年左右在其左臉頰處逐漸發展出一深色且包埋著微小球狀囊腫之班塊。綜合病灶的臨床及組織發現，我們作出粟粒疹板塊的結論並且使用為期3個月的口服美滿黴素治療。病灶幾乎完全消退。此經驗提供我們的資訊指出口服美滿黴素在治療粟粒疹板塊為一種有效且不昂貴的方式。（中華皮誌：26: 180-184, 2008）