A Case of Linear Milia en Plaque on the Central Face

Jung-Wook Kim, M.D., Sang-Hyun Kim, M.D., Doo-Jin Oh, M.D., Deborah Lee, M.D., Seon-Wook Hwang, M.D., Sung-Wook Park, M.D.

Department of Dermatology, Busan Paik Hospital, College of Medicine, Inje University

Milia en plaque (MEP) is a rarely-described benign, cutaneous dermatosis, characterized by numerous tiny milia within an erythematous base which usually occurs around the ears. We report a case of a well-defined, linear MEP arising from the right side of the forehead to the nasal bridge. This case is of particular interest as the site of occurrence is uncommon and the shape of lesion is atypical. (Ann Dermatol (Seoul) 19(2) 81～83, 2007)

Key Words: Linear, Milia en plaque

INTRODUCTION

Milia en plaque (MEP) is a rare clinical variant of milia. The group of milia forming a plaque was first described by Baizer and Fouquet in the retroauricular area in 1903[1]. The term ' ilia en plaque' was given to this atypical clinical form by Hubler et al. in 1978[2]. It arises spontaneously in predisposed individuals without any apparent causative factor[3]. Despite its benign nature and lack of subjective symptoms, MEP raises cosmetic concerns. In most of the previous reports, the lesions were located in the periauricular area[2-6]. We herein report a case of a well-defined, linear MEP arising from the right side of the forehead to the nasal bridge. This developed on an uncommon site and had an atypical shape.

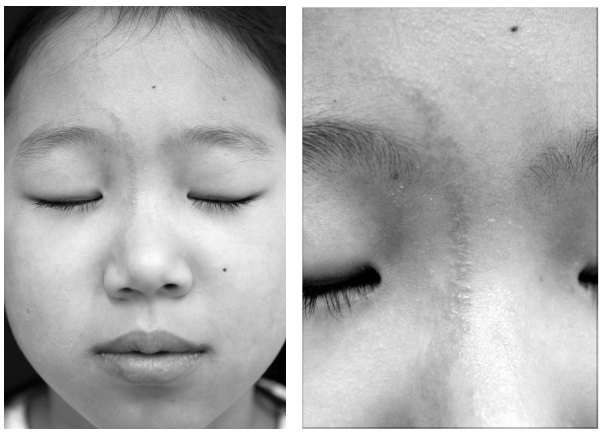

Fig. 1. Multiple, 1 to 2 mm sized, tiny white to yellowish papules were studded on a linear, erythematous plaque from the right side of the forehead to the nasal bridge.

CASE REPORT

A 11 year-old girl presented with multiple, asymptomatic, 1 to 2 mm sized, white and firm papules based on a linear erythematous plaque from the right forehead to the nasal bridge (Fig. 1). The plaque was slightly pinkish in color with a well-circumscribed border. The plaque arose spontaneously on the right side of the forehead and it had progressively spread to the nasal bridge over 8 months. There was no tenderness or local rise of temperature. She did not use any creams, oils, tars, cosmetics, or any other topical agents on the affected area. There was no history of dermabrasion, radiotherapy, burns or trauma. Furthermore, there was no family history of similar disorders. Her general health was excellent. The patient wore glasses but the frames did not rest on the affected area.

Histologic examination of a biopsy specimen obtained from the nasal root revealed a dermal, laminated keratin-filled cyst surrounded by a sparse lymphocytic infiltration compatible with milia (Fig. 2).

Received August 18, 2006
Accepted for publication March 28, 2007

Reprint request to: Deborah Lee, M.D., Department of Dermatology, Busan Paik Hospital, College of Medicine, Inje University, 633-165 Gaegum-dong, Busanjin-gu, Busan 614-735, Korea. Tel: 82-51-890-6135, Fax: 82-51-897-6391, E-mail: medgo@hanmail.net

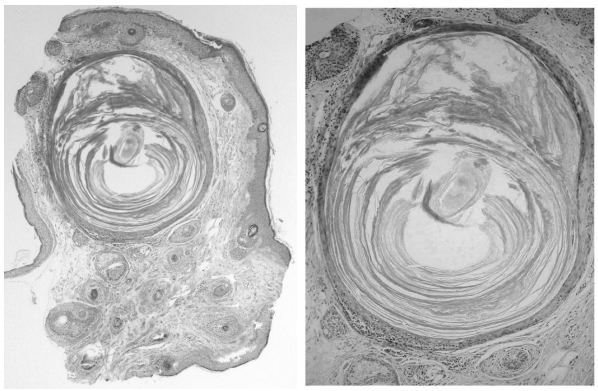

Fig. 2. A keratin-filled cyst is lined by stratified epithelium in the upper dermis and surrounded by sparse lymphocytic infiltration (H & E, × 100).

0.025% tretinoin cream (Stieva-A® 0.025% cream) applied twice daily for 2 months and subsequently 0.25% prednicarbate oint (Dermatop® oint) applied twice daily for 1 month were not helpful. The milia were then manually removed with a 30G needle and comedone extractor. The patient refused any further treatment. The milia were partially eliminated but the linear erythematous plaque remained. There was no significant change to the lesion over the following 12 months.

DISCUSSION

Milia en plaque, a rare subtype of primary milia, is characterized by asymptomatic unilateral or bilateral erythematous, edematous or indurated plaques within which are multiple, whitish-to-yellowish, 1 to 2 mm sized papules. In most of the previous reports, the lesions were located in the periauricular area[2-7]. A few cases have been reported involving other sites, including the eyelid[3,8], inner canthus[3], submandibular area[9], supraclavicular area[10], nasal folds[11], and mental crease[9]. The shape of MEP in the previous reported cases was also mostly irregular. But in our case, the lesion was located from the right side of the forehead to the nasal bridge and the shape of the lesion was linear. MEP usually occurs on adults between the ages of 33 and 65 years (median age 44 years), with a predilection for women (3:1). The average duration prior to diagnosis is 9 months with no familial aggregation noted[3].

Histology of MEP shows typical laminated, keratin-filled, epidermoid cysts circumscribed by mild to dense mononuclear cell infiltrate. However, one case of MEP was characterized histologically by a hybrid cyst with epidermal and trichilemmal keratinization[5]. MEP associated with pseudoxanthoma elasticum[12] and discoid lupus erythematosus[9] have also been reported. In our case, the histologic appearance was compatible with a typical case of milia.

Clinical diagnosis of MEP is not difficult. Differential diagnosis includes Favre-Racouchout disease, familial or naevoid comedo syndromes and lichen planus tumidus follicularis. Milia can be divided into primary and secondary forms. Primary milia develop without a predisposing condition and are most commonly found in adults or during the newborn period. Secondary milia are induced by topically applied perfumes, corticosteroids, 5-fluorouracil, orally administered benoxaprofen, trauma, radiotherapy or blistering diseases[4,7]. And primary milia are believed to be derived from vellus hair, while secondary milia represent retention cysts, usually developed from eccrine sweat ducts or aberrant epidermis of hair follicles[3]. MEP is a rare subtype of primary milia. The distribution of MEP in our case indicated a secondary form. However our case was classified as primary milia, as no known etiologic factors which cause secondary milia formation was identified and there was no evidence of disruption of the pilosebaceous units or sweat structures upon histopathological examination. The pathophysiologic mechanisms which lead to MEP are unknown.

Treatment of MEP is limited and optimal treatment modalities have not yet been established. Simple evacuation is usually inadequate, but in superficially-located milia, incision and expression has resulted in improvement[8]. The response to topical tretinoin used over a period of 3 months varies from a good response[2,10] to no response[6,11]. In cases with dense inflammatory infiltrate of the dermis, minocycline 100 mg/day over 2 to 3 months has been successfully used[7,8]. Surgical treatments such as electrodesiccation and excision have resulted in partial improvement to complete clearance but have been associated with scarring[5,8,13]. Simple spot regional dermabrasion with local anesthesia followed by fusidic acid nonadherent dressing and minocycline 100 mg/day resulted in complete healing without

complication[14]. In our case, topical tretinoin and corticosteroid was unsuccessful and incision and expression proved to be only partially effective.

Our patient was diagnosed with MEP due to clinical appearance and histologic findings. Though the linear distribution of the lesion made us consider a diagnosis of secondary milia, we could not elicit a history of any factor known to induce milia in our case. To the best of our knowledge, our case is the first case of MEP that has involved the right side of the forehead to the nasal bridge with a linear shape. Therefore we report this rare and interesting case of MEP that developed on an uncommon site with an atypical shape. More experience in this rare condition and trial of various treatment modalities is needed.

REFERENCES

1. Balzer F, Fouquet C. Milium confluent retroauriculaires bilateral. Bull Soc Fr Derm Syphilol 1903;14:361.
2. Hubler WR, Rudolph AH, Kelleher RM. Milia en plaque. Cutis 1978;22:67-70.
3. Stefanidou MP, Panayotides JG, Tosca AD. Milia en plaque: a case report and review of the literature. Dermatol Surg 2002;28:291-295.
4. Losada-campa A, De La Torre-Fraga C, Cruces-Prado M. Milia en plaque. Br J Dermatol 1996; 134:970-972.
5. Lee DW, Choi SW, Cho BK. Milia en plaque. J Am Acad Dermatol 1994;31:107.
6. Stork J. Retroauricular bilateral ' ilia en plaque' Dermatology 1995;129:260-261.
7. Keohane SG, Beveridge GW, Benton EC, Cox NH. Milia en plaque-a new site and novel treatment. Clin Exp Dermatol 1996;21:58-60.
8. Bridges AG, Lucky AW, Haney G, et al. Milia en plaque of the eyelids in childhood: case report and review of literature. Pediatr Dermatol 1998;15: 282-284.
9. Kouba DJ, Owens NM, Mimouni D, Klein W, Nousari CH. Milia en plaque: a novel manifestation of chronic cutaneous lupus erythematosus. Br J Dermatol 2003;149:424-426.
10. Combemale P, Faisant M, Dupin M. ' ilia en plaque' n the supraclavicular area. Dermatology 1995;191:262-263.
11. Alsaleh QA, Nanda A, Sharaf A, Al-Sabah H. Milia en plaque: a new site. Int J Dermatol 2000; 39:614-615.
12. Cho SH, Cho BK, Kim CW. Milia en plaque associated with pseudoxanthoma elasticum. J Cutan Pathol 1997;24:61-63.
13. Boehm I, Schupp G, Bauer R. Milia en plaque arising in discoid lupus erythematosus. Br J Dermatol 1997;137:649-651.
14. van Lynden-van Nes AM, der Kinderen DJ. Milia en plaque successfully treated by dermabrasion. Dermatol Surg 2005;31:1359-1362.