Multiple milia formation following refractory bullous pemphigoid

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Received May 11, 2015; Accepted May 25, 2015; Published Aug 28, 2015

ABSTRACT

Formation of milium after bullous pemphigoid (BP) is not so rare, but the cases presenting numerous number of milia like our case is rare. We report a 70-year-old male who developed a remarkable number a milium after BP improved. The number of milium in our case might be more than previous reports. One of the cause of a remarkable number of milium appeared in our case might be his chronic clinical course. In his 6 months of hospitalization, erythema, blister and erosion relapsed many times over. Every time erosions cured, keratin might be entrapped, and it might lead a remarkable number of milia.

CASE REPORT

A 70-year-old male was referred to our department, complaining of generalized blisters and erosions. He had been suspected of bullous pemphigoid (BP) and treated with oral prednisolone (PSL) (30 mg/day), however, his eruptions got worsened. Physical examination revealed erythema and a number of scattered, tense blisters and erosions on the trunk and extremities (Figure 1). Laboratory data by ELISA showed elevated anti-BP180 antibody titer (2040; normal < 9). A biopsy specimen taken from blister showed subepidermal bulla, and inflammatory cell infiltration, mainly composed of eosinophils (Figure 2). Direct immunofluorescence showed linear deposit of IgG in epidermal side of dermo-epidermal junction (Figure 3). We treated him with immunosuppressant (ciclosporin and methotrexate), and plasma exchange in addition to oral PSL. Erosions and blisters were slowly cured. After erosions were cured, a number of milium-like eruptions that was more than 100 appeared scattered on trunk and extremities (Figure 4, 5). A biopsy specimen showed a cystic nodule with epithelial wall. The border of nodules was clear, and lumen of the cyst was filled with keratin (Figure 6).

DISCUSSION

Milium is a common skin cyst which is formed at the base of a hair follicle or sweat gland, and could be categorized as primary or secondary. Primary milia are formed from entrapped keratin, and are usually found on the faces. Secondary milium develops after an injury, burn, skin graft, or bullous diseases such as BP [1]. One of the treatment for milium is to puncture it and pressing out the keratin inside the milium.

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Figure 2. Histopathological specimen, shown by Hematoxylin-eosin stain, showed subepidermal bulla, and inflammatory cell infiltration, mainly composed of eosinophils.

Figure 3. Direct immunofluorescence showed linear deposit of IgG in epidermal side of dermo-epidermal junction.

Figure 4, 5. A numbers of tiny whitish eruptions appeared scattered on the trunk and extremities (hands: Figure 4, arms: Figure 5).

Figure 6. A biopsy specimen showed a cystic nodule with epithelial wall. The border of nodules was clear, and lumen of the cyst was filled with keratin.
Formation of milium after BP is not so rare, but the cases presenting numerous number of milia like our case is rare. In a recent literature, Tsuruta et al. reported multiple large milia appeared on BP patients [2]. In our case, remarkable number of milium appeared, and it might be more than previous reports. One of the cause of remarkable number of milium appeared in our case might be his chronic clinical course. In his 6 months of hospitalization, erythema, blister and erosion relapsed many times over. Every time erosions cured, keratin might be entrapped, and it might lead remarkable number of milia. However there may be other causes of multiple milia formation, and further studies are necessary.

REFERENCES
