CORRESPONDENCE

Systemic isotretinoin with topical tretinoin cream in the treatment of multiple warty dyskeratomas

Case report

A 29-year-old Taiwanese man initially presented with a painful red nodule on the right cheek for several days. A total excision was arranged. Histopathologically, the lesion demonstrated features of warty dyskeratoma (WD) characterized by a dilated cystic epidermal invagination with a hyperkeratotic and parakeratotic plug. Suprabasal acantholysis was prominent, as were acantholytic and dyskeratotic cells, corps ronds, and grains. He came to our clinic 2 years later because more lesions appeared on his face and trunk gradually over these years. Physical examination at that point revealed hundreds of keratotic papules or nodules with follicular plugging

Figure 1  Multiple warty dyskeratomas characterized by erythematous papules, nodules with umbilicated or a pore-like keratotic center on the (A) face and (B) scalp. (C and D) The size and number of the lesions improved dramatically after 1 month of oral isotretinoin combined with topical 0.05% tretinoin cream.
on the scalp, face, neck, and the chest (Figure 1A and B). Another skin biopsy from a scalp lesion was performed and microscopically showed similar pathological findings as the previous one, a typical WD (Figure 2). He had no personal or family history of Darier’s disease or other skin disorders. He was treated with oral isotretinoin 30 mg/d in three divided doses along with topical 0.05% tretinoin cream. Both the size and number of the lesions improved significantly after 1 month of therapy (Figure 1C and D). Mild dry mouth was the only adverse reaction been complained. The patient stopped the treatment 14 weeks later because of the cost and the side effect. In the 5 months follow-up after discontinuing the medications, most of the lesions have remained as small as they were during treatment, and only a few new ones have developed (Figure 3).

Discussion

The report provides a successful experience of medical treatment of WD. WD most commonly presents as a single nodule and locates on the areas of head and neck. Accordingly, diagnosis and treatment can be achieved concurrently by excisional surgery. However, multiple WDs are rarely documented and just six case reports have been published, which are summarized in Table 1. There is little information about the medical treatment of multiple WDs; only one case reported by Abramovits and Abdelmalek showed excellent response by topical tazarotenic acid in their case.

There are some similarities between the pathogenesis of acne and that of WDs. The major elements of acne formation include infundibular epidermal hyperproliferation, excess sebum production, inflammation, and the presence of activated Propionibacterium acnes. On the other hand, most cases of WDs show variable signs of differentiation toward the follicular infudibulum, which includes the presence of multiple small infundibular cystic structures, and of focal contiguity to pilosebaceous structures. Systemic isotretinoin has been used over the past 2 decades for treating severe nodular acnes by reduction in size and output of sebaceous glands, as well as modulation in keratinocyte maturation. Isotretinoin is presumed effective for multiple WDs for that it can be seen as acneiform eruptions. In addition, systemic isotretinoin has been well known for its good effects to epidermal proliferation and keratinizing process. Immunomodulatory, anti-inflammatory, anti-cancer, and antiangiogenic effects have also been documented. Therefore, isotretinoin has been used for the treatment of several non-acne dermatologic diseases, such as psoriasis, pityriasis rubra pilaris, Darier’s disease, skin cancers, and so forth. WD is defined as...
a disorder of epidermal proliferation and the multiple ones share some features with acne vulgaris and Darier’s disease. As a result, systemic isotretinoin is theoretically the treatment of choice.

Compared with the patient reported by Abramovits and Abdelmalek, who had grouped small follicular papules locating restrictively over the left groin area, the lesions in our case are clinically more extensive in distribution (scalp, face, neck, and chest vs. left groin), and the individual nodule is larger in size (nodule vs. papule). Therefore, the treatment with topical retinoid seems insufficient for such widely distributed lesions. The result of combination of systemic isotretinoin with topical tretinoin cream gives a marked improvement that lasts for at least 5 months even after the treatment is discontinued.

This case demonstrates a dramatic response of multiple WDs to oral isotretinoin with topical tretinoin cream in the 1st month of treatment. The efficacy of the treatment lasts for at least 5 months even after the treatment is discontinued.

Ongoing follow-up will be needed to see if the effect is maintained.

Ying-Ling Kuo
Department of Dermatology, Mackay Memorial Hospital,
Taipei, Taiwan

Yu-Hung Wu∗
Department of Dermatology, Mackay Memorial Hospital,
Taipei, Taiwan

Mackay Medicine, Nursing and Management College,
Taipei, Taiwan

Ying-Jui Chang
Department of Dermatology, Far Eastern Memorial Hospital,
Taipei, Taiwan

∗ Corresponding author. Department of Dermatology, Mackay Memorial Hospital,
No. 92, Section 2, Chung-Shan North Road,
Taipei 104, Taiwan.
E-mail address: yhwu@ms2.mmh.org.tw (Y.-H. Wu).

Table 1 Summary of six case reports of multiple warty dyskeratomas.

<table>
<thead>
<tr>
<th>Year (ref.)</th>
<th>Age/sex</th>
<th>Race</th>
<th>Underlying disease</th>
<th>Lesion number/size (mm)</th>
<th>Location</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>19871,2</td>
<td>NA</td>
<td>Japanese</td>
<td>Renal dysfunction</td>
<td>3–5/NA</td>
<td>Head and neck</td>
<td>Not mentioned</td>
</tr>
<tr>
<td>19933</td>
<td>63/M</td>
<td>Japanese</td>
<td>Nephrotic syndrome</td>
<td>NA/5–7</td>
<td>Scalp, face, neck, and hands</td>
<td>Not mentioned</td>
</tr>
<tr>
<td>19974</td>
<td>64/F</td>
<td>Caucasian</td>
<td>Dysrhythmia</td>
<td>25/8–10</td>
<td>Scalp</td>
<td>Not mentioned</td>
</tr>
<tr>
<td>1993</td>
<td>84/F</td>
<td>African American</td>
<td>Hypertension</td>
<td>15/3–5</td>
<td>Scalp</td>
<td>Not mentioned</td>
</tr>
<tr>
<td>20025</td>
<td>27/F</td>
<td>Caucasian</td>
<td>None</td>
<td>NA/NA</td>
<td>Left groin</td>
<td>Topical retinoid</td>
</tr>
<tr>
<td>20096</td>
<td>53/F</td>
<td>NA</td>
<td>None</td>
<td>15/5–13</td>
<td>Scalp</td>
<td>Surgically excised</td>
</tr>
<tr>
<td>Our case</td>
<td>29/M</td>
<td>Taiwanese</td>
<td>Chronic liver disease</td>
<td>240 on scalp, 50 on other areas/4–8</td>
<td>Scalp, face, neck, and chest</td>
<td>Oral and topical retinoids</td>
</tr>
</tbody>
</table>

References