

A Case of Multiple Keratoacanthomas Associated with Keratoacanthoma Centrifugum Marginatum

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Accepted September 12, 2005.

Sir,

Keratoacanthoma (KA) is a benign cutaneous neoplasm that resembles squamous cell carcinoma both clinically and histopathologically. It is usually characterized by rapid onset and spontaneous regression (1–3). Although solitary type KA is the most common, there have been rare reports of both keratoacanthoma centrifugum marginatum (KCM) type and multiple KAs. Development of multiple KAs and KCM in a single patient has been reported previously in three cases (3).

CASE REPORT

A 45-year-old man attended our clinic with multiple asymptomatic papules and nodules on his gluteus, lower extremities and right hand. The lesions first appeared on his right hand and knees 6 years previously and spread to his gluteus and lower extremities. Some of the lesions regressed spontaneously leaving atrophic scars. There was no family history of any type of KA and no personal or family history of malignancy. He had not used immunosuppressive drugs and had no contact with any chemicals. Dermatological examination revealed multiple (approximately 50) 0.3–3 cm, bluish-red, crater-like lesions on his gluteus and lower extremities (Fig. 1). There was a 10×10-cm annular eruption with hyperkeratotic, pinkish-brown margins that had a tendency to expand peripherally on the dorsum of his right hand (Fig. 2). Routine laboratory tests were normal. Serology for human immunodeficiency



Fig. 1. Multiple, 0.3–3 cm, bluish-red, crater-like lesions on the patient's gluteus. The prominent scars are due to previously made excisional biopsies.



Fig. 2. Annular eruption with hyperkeratotic, pinkish-brown margins that had a tendency to expand peripherally on the dorsum of the patient's right hand.

virus (HIV) was negative. Physical and laboratory examinations revealed no internal organ malignancy. Histopathological examination of multiple biopsy materials revealed squamous proliferative lesion with a central irregular crater filled with keratin. There were horn cysts at the base of the crater (Fig. 3). The patient

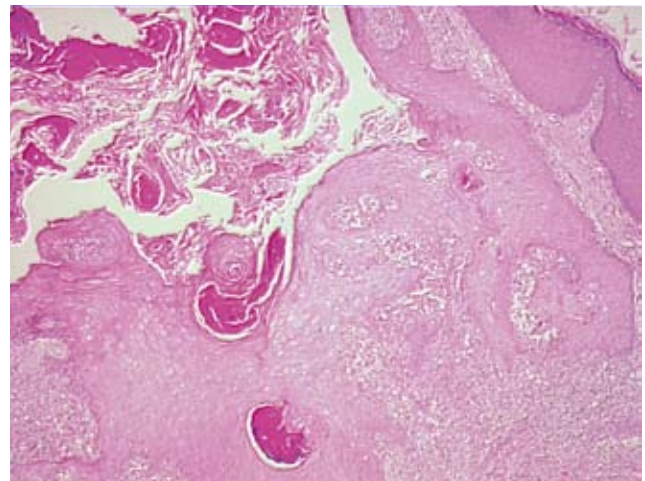


Fig. 3. Squamous proliferative lesion with a central irregular crater filled with keratin. The epidermis extends over the sides of the crater. There are horn cysts at the base of the crater. An inflammatory infiltration is present in the dermis, just beneath the base of the lesion (haematoxylin-eosin ×40).

was diagnosed as having multiple KAs and KCM with clinical and histopathological findings.

DISCUSSION

Several types of KA have been described. The most common type are solitary, but there are also multiple KAs known as Ferguson-Smith type KAs, Grzybowski type KAs, Witten and Zak type and Muir-Torre syndrome (1, 3–6). The classification of multiple KAs is not standardized and often leads to confusion and overlapping (3). Ferguson-Smith type KA usually begins in childhood or adolescence and appears to be inherited in an autosomally dominant pattern (1). In this type, there are fewer lesions and they have a tendency to ulcerate and heal spontaneously after persisting for a long time, leaving atrophic scars (3, 6). Generalized eruptive KAs of Grzybowski appear as sporadic, disseminated 2–3 mm KAs most commonly affecting patients between 50 and 80 years of age. The response to therapy is poor (7). The Witten and Zak type is a coexistence of these two types (6).

Association of multiple KAs with carcinomas has been described by a number of authors. Multiple KAs and sebaceous neoplasms associated with internal malignancy is named Muir-Torre syndrome (3).

The cause of solitary or multiple KAs is unknown. Heredity, viruses, chemicals and sunlight are thought to be involved in the aetiology of KAs. The incidence of KAs has been reported to be increased in patients who have a history of prolonged contact with insecticides, printing chemicals, tar or pitch (8–10). KAs may be seen in association with malignancies, xeroderma pigmentosum and immunosuppression. Most of our patient's lesions were located on skin that had not been exposed to sunlight. There was no history of contact with any chemicals. We did not detect any malignancy in the clinical and laboratory examinations.

KCM is an uncommon and distinct type of KA with central healing and progressive peripheral expansion. KCM may rarely be multiple (1). Multiple KAs associated with KCM is extremely rare. Schaller et al. (3) reported a case with multiple KAs, giant KA and KCM. Their case was Ferguson-Smith type because the patient had family history of KAs. Another case that had eruptive KAs of Grzybowski, KCM and a solitary, typical nodular keratoacanthoma was reported from Poland (11). Kato et al. (12) reported a Japanese patient

with both Ferguson-Smith type KAs and KCM in 2003. Our case is the fourth reported case of a combination of multiple KAs and a KCM.

Our case shows some characteristic clinical features of Ferguson-Smith type (few lesions, larger formation and healing with scars), but there was no family history of KAs and it began at an older age. Our patient may represent a case of multiple KAs of the Ferguson-Smith type, which developed with genetic mutations, because there was no family history of KAs.

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