Multiple Basal Cell Carcinoma Associated with Keratoacanthoma

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We report a case of multiple basal cell carcinoma associated with keratoacanthoma. A 65-year-old Korean female had suffered from multiple, variable-sized papules and nodules on the face for 20 years prior to treatment. She had no history of arsenic intake, irradiation, herb medication, or hereditary or preexisting dermatoses. Histopathologically, the tumors revealed typical findings of solid and adenoid types of basal cell carcinoma and keratoacanthoma.

Key Words: Multiple basal cell carcinomas, keratoacanthoma

Basal cell carcinoma (BCC) usually occurs as a single lesion, mainly on sun-exposed areas, although the occurrence of several lesions is not unusual (Lasser and Carter 1973; Williamson et al. 1967; Murray and Cannon 1960). Multiple BCC may be associated with arsenic exposure, in which case it occurs commonly on the neck, trunk and back, and with familial predisposing diseases such as nevoid basal cell epithelioma syndrome, Bazex syndrome and xeroderma pigmentosa (Southwick and Schwart 1979; Holubar et al. 1970; Fitzpatrick et al. 1987). However, the onset of the latter conditions usually begins in childhood. To our knowledge, the association of BCC and keratoacanthoma is not previously mentioned in the literature.

CASE REPORT

A 65-year-old Korean woman whose skin color was type III, was seen by us in June of 1990 with a complaint of multiple, variable-sized, symptomless papules and nodules that had gradually appeared on the forehead, nasolabial folds and right cheek for approximately 20 years (Fig. 1A). A 1 cm-sized hard, single, dome-shaped nodule had suddenly developed on the left mandibular area about one year previous to our examination (Fig. 1B). The largest ulcerative lesion on the right cheek recently and rapidly grew in size with central hemorrhagic necrosis. There were no regional lymphadenopathies. Familial and past history were noncontributory.

CBC, urinalysis, liver function test, chest X-ray, VDRL and EKG were within normal limits or negative. The whole body bone scan for the evaluation of metastasis revealed hot uptakes in the lumbar spine and hip bone area. Skin biopsies of the pigmented tumor on the forehead and a large ulcerative tumor on the right cheek exhibited the adenoid and solid type of BCC, respectively (Fig. 2A, B). A biopsy obtained from the left mandibular area revealed typical findings of keratoacanthoma showing the large central keratin-filled crater, the epidermal
Fig. 1. A: Pea to thumb-tip sized, peripherally rolled bordered, or slightly ulcerative tumors on the forehead and nasolabial folds, and 3 x 4 cm sized, hard, fixed, and ulcerative tumor on the right cheek.
B: 1 x 1 cm sized, hyperkeratotic, dome shaped, hard, fixed tumor on the left mandibular area.

Fig. 2. A: Adenid type of BCC from the tumor on the forehead (H & E, x 100).
B: Solid type of BCC from the large ulcerative tumor on the right cheek (H & E, x 100).
C: Large, central, keratin-filled crater showing the typical shape of keratoacanthoma, from the tumor on the left mandibular area (H & E, x 20).
extensions over the side of crater and epidermal proliferations extending downward from the base of crater into the dermis. The tumor cells appeared glassy, and there were several horn pearls. However, atypical cells were not observed (Fig. 2C).

The tumors other than the ulcerative lesion were completely removed with excisional biopsies and electrocauterization. Excisional biopsies of the erosive surfaced tumor on the forehead revealed morphea type, and the peripherally rolled bordered tumor on the left inner canthus and the pigmented tumor on the left nasolabial fold exhibited adenoid types of BCC. The tumors being removed by electrocauterization were not observed microscopically.

The patient refused further treatment of the main ulcerative mass. Six months after her first visit she died. No autopsy was performed.

**DISCUSSION**

BCC is the most common form of skin cancer, accounting for 65% to 75% of all cutaneous tumors (Wermuth and Fajardo 1970). Although the occurrence of multiple lesions either simultaneously or subsequently is not uncommon, BCC usually occurs as a single lesion, mainly on the face and neck, with a small percentage arising on an extremity.

BCC arises from the basal cell of the epidermis and from the external root sheath of hair follicles. The actual cause of BCC is unclear, but the role of sunlight in the pathogenesis of BCC and keratoacanthoma is well established (Fitzpatrick et al. 1987). Multiple BCC may be associated with arsenic intake, burn scar, or hereditary diseases such as xeroderma pigmentosa, Bazex syndrome, nevoid basal cell epithelioma syndrome, and albinism (Southwick and Schwartz 1979; Holubar et al. 1970; Fitzpatrick et al. 1987; Wagner et al. 1979; Marvin 1970).

Our case, however, had none of those conditions. With regard to the subsequent occurrence of BCC and keratoacanthoma, we cannot completely rule out chronic sun exposure as a possible cause. Because we could observe the some degree of solar degenerative changes in the upper dermis of the some biopsy specimens of BCC and a keratoacanthoma, and the patient had worked at the farm for a long time, she might had been damaged her skin from sunlight.

There are several clinical and histologic types of BCC. Distinctive dividing criteria between these histologic types can not be easily determined, because many lesions showed admixtures of the various types. Most of our specimens showed solid and adenoid patterns histologically and fibrosing BCC clinically (Lever and Schauburg-Lever 1990).

In spite of the frequency of BCC and its capacity for local destruction, metastasis is distinctly unusual. The recorded incidence in a number of large series ranges from 0.0028% to 0.55% (Farmer and Helwig 1980; Domarus and Stevens 1984). About 170 documented cases have been reported in recent English language review articles.

BCC usually metastasizes to regional lymph nodes, followed by lung, liver, bone, spleen and adrenal glands (Farmer and Helwig 1980; Domarus and Stevens 1984; Safai and Good 1977; Binkley and Rauschkolb 1962; Assor 1967; Mikhail et al. 1977). Clearly, metastasis can occur by either the lymphatic or hematogenous routes. The interval between the onset of the primary tumor and the metastatic lesion varied in reports from 0 to 30 years. Mean survival time after metastasis is approximately 10 months (Farmer and Helwig 1980).

Lattes; Kessler (1951), Cotran (1961), and Wermuth and Fajardo (1970) have established the criteria for diagnosing metastasis from BCC. Although we did not perform a bone biopsy, we highly suspected metastasis to the bone. First of all, the whole body bone scan revealed hot uptakes at the site of the lumber, and sacral spine and hip bone. Secondly, the patient expired after 6 months. Lastly, the majority of metastatic BCC originates from chronic, large, ulcerative lesions. Our case had a huge, hemorrhagic, necrotic ulceration on the left cheek area.

Close observation of multiple, recurrent, long standing and large ulcerative lesions will be mandatory for early detection of metastasis and proper management.

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