Hypercalcemia associated with squamous cell carcinoma arising in pyoderma chronica

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LETTER TO THE EDITOR

Hypercalcemia associated with squamous cell carcinoma arising in pyoderma chronica.

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Dear Editor

A 59-year-old man had a 43-year history of pyoderma chronica involving buttocks, nape, back, chest, and abdomen. He developed an ulcerated tumor on his left scapular area in April 2006. Physical examinations revealed a $7 \times 5$ cm reddish ulcerative tumor on his left scapular region (Fig.1). An incisional biopsy disclosed well differentiated squamous cell carcinoma. He also had multiple bilateral axillary lymphadenopathies. Magnetic resonance imaging showed extension to the subcutaneous tissue, but not to the underlying muscle. A total body computed tomography (CT) scanning demonstrated no distant metastasis. Routine laboratory studies showed the white cell count was 15,860/ mm$^2$, C-reactive protein (CRP) was 1.99 mg/dl. Liver function and renal function tests were within normal ranges. SCC antigen was elevated to 5.4 ng/ ml (normal: <1.5 ng/ml). A wide local excision with 2-cm margin deep into the trapezius fasciae
was performed. Bilateral axillary lymph node biopsy revealed left node involvement, which was dissected. Histopathologic examination of the tumor revealed well differentiated SCC, with tumor nests extending to the deep dermis and subcutaneous tissues. A sinus tract was lined by atypical tumor cells (Fig. 2). All margins were free of the tumor cells.

Nine months later, however, he complained pain on his left scapular area. A CT scanning showed low density area in trapezius muscle. Biopsy revealed poorly differentiated SCC. At this time, decreased consciousness was noted and laboratory test disclosed hypercalcemia. Serum level of calcium increased to 12.3 mg/dl, serum phosphorus decreased to 1.6 mg/dl, parathyroid hormone level was 5 pg/ml (normal: 10-65 pg/ml), and parathyroid hormone-related peptide (PTHrP) was 3.6 pmol/l (normal<1.1 pmol/l). SCC antigen was markedly elevated to 22.3 ng/ ml. He was managed with hydration and
parenteral bisphosphonate infusions to control hypercalcemia. A total CT scanning demonstrated no distant metastasis. Bone scanning showed significantly increased uptake on his left scapula. He was treated with combination chemotherapy of cisplatin and adriamycin, followed by local radiation therapy on the recurrent site. The size of tumor mass was decreased and hypercalcemia was improved. SCC antigen decreased to 4.8 ng/ml and PTH-rP level reduced to 1.0 pmol/l. However, he developed multiple lung and bone metastases, and died 18 months after the initiation of the therapy.

Pyoderma chronica, synonymous to acne conglobata and hidradenitis supprativa, is a chronic suppurative scarring disease which typically involves buttocks, perianal and anogenital regions. Malignant transformation of pyoderma chronica is rarely observed\(^1\), \(^2\). It tends to be more locally aggressive than the usual cutaneous SCC with higher incidence of distant
metastasis and poor prognosis. Maclean and Coleman\textsuperscript{3} reported, 15 out of 31 cases of pyoderma chronica-related SCC died within 2 years following the diagnosis of SCC. Our patient who had a positive lymph node metastasis died in 18 months following the initial treatment. Regional lymph node metastasis in pyoderma chronica-related SCC is known to be a poor prognostic sign. So far 14 detailed description is available in Japan and among these, 9 died with average survival of 7 months\textsuperscript{4}. The poor prognosis is related to advanced stage at the time of diagnosis, because the SCC usually invades deep in the fistules.

Malignancy-associated hypercalcemia is occasionally observed. One is humoral hypercalcemia of malignancy and the other is local osteolytic hypercalcemia and the former consists of 80%. The prognosis of hypercalcemia is also poor; approximately 75% of cases died within 3 months following the treatment\textsuperscript{5}. There are 10 reports of
humoral hypercalcemia of malignancy in association with primary cutaneous SCC\textsuperscript{2,6-9}. Serum PTH-rP was increased in 7 cases examined. Among them, 3 patients died within 1 month and 6 died within 3 months. However, in 3 cases tumor excision improved hypercalcemia with decrease in serum PTH-rP level\textsuperscript{6,7,8}. In 1 case complicated with pyoderma chronica, calcium level was well controlled by chemotherapy\textsuperscript{2}.

In our case, because increased serum PTH-rP and hypercalcemia with concomitant decrease in serum PTH and phosphorus levels was improved after the combination therapy temporarily, the major cause of hypercalcemia is most-likely due to the PTH-rP secreted by the tumor.
References


Figure 1. A $7 \times 5$ cm reddish ulcerative tumor on the left scapular region

Figure 2. A sinus tract was lined by atypical tumor cells