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— Abstract —

Surgical Treatment of Squamous cell Carcinomas Arising in Scalp Burn Wounds - Two Case Reports -

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Marjolin's ulcer is a rare and often-aggressive cutaneous malignancy that arises in previously traumatized or chronically inflamed skin, particularly after burns. We experienced two cases after burns.

Case I involved a forty eight year-old man who had suffered from a flame burn at the parietal scalp area, where had been initially described three years earlier as a full-thickness wound including the pericranium. The man consulted us for a persistent ulcerative and infected wound on the burned lesion during the last 24 months, which turned out on the contrast-enhanced computed tomography (CT) and magnetic resonance imaging (MRI) to be the squamous cell carcinoma with involving the skull and the dura mater. Although the posterior auricular lymph node was enlarged on the ipsilateral side, recent positron emission tomography (PET) CT did not show any metastatic lesion. It was impossible for us to resect the intracranial involvement of the tumor radically, and the postoperative PET CT still showed a focal fluorodeoxyglucose (FDG) uptake around the wall of the superior sagittal sinus. We think that an aggressive combined approach is essential for treatment in early stages for a high success rate, before the intracranial structures are involved because there is no consensus on the treatment for advanced disease, and the results are generally poor. Case 1 also did not involve a radical resection because of the intracranial invasion to the wall of superior sagittal sinus and the possibility of damage to the major cortical veins. He received adjuvant radiotherapy and must be followed periodically.

Case 2 involved an eighty six year-old women who suffered from a painful scalp ulcer lesion after flame burns three years earlier. Unlike case 1, neither tumor infiltration into the dura nor lymph node enlargement was observed on the contrast-enhanced computed tomography (CT), magnetic resonance imaging (MRI), or positron emission tomography (PET) CT. We did a radical resection of the tumor, including the involved bone, and a cranioplasty with bone cement. (J Korean Soc Traumatol 2007;20:52-56)

Key Words: Marjolin's ulcer, Squamous cell carcinoma, Burn, Scalp

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I. Introduction

Marjolin's ulcer on the scalp is uncommon condition in which malignant transformation occurs in chronically inflamed or traumatized skin, it is first reported by J.N. Marjolin in patients who developed malignant ulcer from burn scars in 1828.(9,13) Treves and Pack(13) estimated that about 2% of burn scars undergo malignant change and they found that these usually occurred in the extremities, trunk, and scalp. Squamous cell carcinoma (SCC) is the major histological type of Marjolin's ulcer.(1,3,6,8,12,14) We experienced the treatment of Marjolin's ulcer of the scalp with intracranial involvement.

II. Case report

1. Case 1

A 48-year-old man was transferred to our clinic with a $9 \times 7 \times 3$ cm central ulcerating mass on the scalp (Fig. 1). He was burned with flame 45 years ago. He had no problem except the bald area for years, until a bleeding wound occurred in the center of the bald area 6 months before his

admission. The scalp wound was a full-thickness burn scar including pericranium.

Contrast-enhanced magnetic resonance image (MRI) of the head revealed that scalp mass involved the skull and dura mater at the skull vertex. Positron emission tomography computed tomography (PET CT) demonstrated FDG uptake of the lesion but did not show any metastatic lesion



Fig. 1. The patient's picture shows a huge inflamed ulcerative lesion on scalp.



Fig. 2. (A) MRI shows $6.9 \times 6.4 \times 3.1$ cm sized soft tissue mass, which involves the skull and dura. (B) Preoperative PET-CT shows intense FDG uptake on the lesion. The enlargement of lymph node suggests inflammatory condition (white arrow: lymph node).

(Fig. 2). The operation was started with 3 cm clear margins down to the parietal bones, which was considered to be tumor-free. As the wall of superior sagittal sinus was involved, we tried to peel and desiccate the layer of dura including tumor tissue as much as possible. The defect of surgical wound was reconstructed by the hydroxyapetite cement and a split-thickness skin graft over the latissimus dorsi myofascial free flap (Fig. 3). Histopathologic finding of mass revealed SCC, which was well differentiation with keratinization. Immunohistochemical stain of mass was positive for keratin, but nonreactive for Vimentin and S-100 protein (Fig. 4).

He healed well without any complication, and adjuvant radiation therapy was performed with 6290 cGy because



Fig. 3. The patient has a clean and good circulated flap on the parietal scalp.

the FDG uptake was still remained at the dura around superior sagittal sinus, and shown at left cervical lymph node which suggested inflammatory reaction on PET CT. After adjuvant radiation therapy, a focal FDG uptake around superior sagittal sinus and left cervical lymph node disappeared on follow-up PET CT (Fig. 5).

2. Case 2

A 86-year-old women also had suffered from painful scalp ulcer lesion after flame burns at 3 year-old. Unlike case 1, there were no tumor infiltration into dura and lymph node enlargement on the contrast-enhanced CT, MRI and PET CT. We performed wide excision, including tumor and bone and then cranioplasty was done with bone cement.

III. Discussion

Marjolin's ulcers are rare tumors that originate in chronically inflamed skin. The inflammation is usually caused by a burn injury. However, vaccination scars, snakebite scars, osteomyelitic scars, chronic pilonoidal abscesses, pressure sores and venous stasis ulcers may also induce this tumor.(1,3,6,7,11,12,14) Risk factors for cutaneous squamous cell carcinoma include long-term exposure to ultraviolet, ionizing radiation or arsenic and repeated contact with polycyclic hydrocarbons.(1,6,12) The mechanism of malignant change is supposed to be a sequence of repeated ulceration, trauma and healing in the ulcers and osteomyelitic sinuses. Some proposed mechanisms include carcinogenic toxins pro-



Fig. 4. (A, B) (Micro, H-E stain, ×40 and ×200): It is composed of larger cells with hyperchromatic pleomorphic nuclei around the stroma infiltrated with lymphocytes and plasma cells.

duced from the burned tissue, the cicatricial tissue that prevents the immunologic mechanisms from checking the new tumor formation.(2,12,13) The incidence of this disease is very low. Traves and Pack(13) reported that among 2465 cases of squamous cell and basal cell carcinomas, 28 cases were due to carcinoma arising from burn scars. These lesions most commonly show the histopathology of SCC, although cases of basal cell carcinoma, melanoma, malignant fibrous histiocytoma, liposarcoma, and fibrosarcoma have also been reported.(1,3,8,10,12-14)

Marjolin's ulcer is divided into 2 categories: acute type, arising within 12 months of the burn, and the more common chronic type, arising any time after 12 months. In the former the average latency is 4 weeks to 1 year, whereas in the latter the average period is 36 (range, 1-75) years. (1,7,12,14) There is always a latent period between burn injury and appearance of the turnor, and average lag period is 30 years, although an acute onset may occur rarely, sometimes within weeks(3,14). In our series, the lag periods were 45 years and 83 years, respectively.

The carcinoma developed from burn scar has a propensity for the extremities, specifically to flexion creases of the extremities, because blood supply of the extremities is decreased and trauma is increased. The role of diminished blood supply may explain why the Marjolin's ulcers on the scalp have a relatively low incidence, because the scalp have a high vascularization, (5,12) The most important prognostic factors are pathologic type, grade of the lesion and lymph node metastasis at the time of the diagnosis, (1,6,11,12) It may easily be predicated that poorly differentiated SCC have a tendency to spread to lymph nodes earlier. (1,3,4,6,11) Most studies have not supported the idea of prophylactic lymph node dissection like this case, (3)although Novick (10) et al suggested that prophylactic lymph node dissection should be performed for tumors located on the lower extremities.

Complete excision of cancer and skin coverage with a flap or graft is the treatment of choice for the carcinoma developed from burn scar. Wide excision with a surgical margin at least 2 cm is suggested.(3,6,7,11,12,14) It has been recommended that regional lymph node dissection be performed only in the presence of clinical-histologic positive nodes and not as a prophylactic measure.(1,6,11,12) In conclusion, in case of chronic burn scars over the scalp, periodic observation is essential to avoid the morbidity and mortality by malignant transformation with intracranial involvement and metastasis. PET CT is helpful to avoid radical neck dissection because it can discriminate between the reactive inflammatory lymphadenopathy by Marjolin's ulcer and metastatic lymphadenopathy.



Fig. 5. (A, B) PET CT has still a focal FDG uptake on the dura layer around superior sagittal sinus, which suggests residual tumor (Black & White arrow). After radiation therapy, a focal FDG uptake around superior sagittal sinus and left cervical lymph node disappeared. (C) On post operative state, left neck lymph node decreased in size without FDG uptake (Black arrow head). After radiation therapy, left cervical lymph node disappeared.

REFERENCES

- Aydogdu E, Yildirim S, Akoz T: Is surgery an effective and adequate treatment in advanced Marjolin's ulcer? Burns 2005:31:421-31.
- Bostwick J, 3rd, Pendergrast WJ Jr, Vasconez LO: Marjolin's ulcer: an immunologically privileged tumor? Plast Reconstr Surg 1976;57:66-9.
- Copcu E, Aktas A, Sisman N, Oztan Y: Thirty-one cases of Marjolin's ulcer. Clin Exp Dermatol 2003; 28:138-41.
- 4) Eastman AL, Erdman WA, Lindberg GM, Hunt JL, Purdue GF, Fleming JB: Sentinel lymph node biopsy identifies occult nodal metastases in patients with Marjolin's ulcer. J Burn Care Rehabil 2004:25:241-5.
- Fishman JR, Parker MG: Malignancy and chronic wounds: Marjolin's ulcer. J Burn Care Rehabil 1991; 12:218-23.
- Gul U, Kilic A: Squamous cell carcinoma developing on burn scar. Ann Plast Surg 2006:56:406-8.
- Horch RE, Stark GB, Beier JP: Unusual explosive growth of a squamous cell carcinoma of the scalp after electrical burn injury and subsequent coverage by

sequential free flap vascular connection--a case report. BMC Cancer 2005:5:150.

- Love RL, Breidahl AF: Acute squamous cell carcinoma arising within a recent burn scar in a 14-year-old boy. Plast Reconstr Surg 2000;106:1069-71.
- Marjolin JN: Ulcere Dictionnaire de Medicine. Paris, France Vol 21, 1828.
- 10) Novick M, Gard DA, Hardy SB, Spira M: Burn scar carcinoma: a review and analysis of 46 cases. J Trauma 1977:17:809-17.
- Ozek C, Cankayali R, Bilkay U, Guner U, Gundogan H, Songur E, et al: Marjolin's ulcers arising in burn scars. J Burn Care Rehabil 2001:22:384-9.
- 12) Ozek C, Celik N, Bilkay U, Akalin T, Erdem O, Cagdas A: Marjolin's ulcer of the scalp: report of 5 cases and review of the literature. J Burn Care Rehabil 2001:22:65-9.
- 13) Treves N, Pack GT: The development of cancer in burn scars: An analysis and report of 34 cases. Surg Gynecol Obstet 1930;51:749-82.
- 14) Wong A, Johns MM, Teknos TN: Marjolin's ulcer arising in a previously grafted burn of the scalp. Otolaryngol Head Neck Surg 2003;128:915-6.