



CASE REPORT

Invasive Cutaneous Squamous Cell Carcinoma Arising from Chronic Hidradenitis Suppurativa: A Case Report of Treatment by Slow Mohs Micrographic Surgery

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Hidradenitis suppurativa (HS) is a chronic recurrent inflammatory condition presenting with painful, deep-seated abscesses and sinus tracts in multifocal locations. Rarely, long-standing inflammation in HS may lead to serious complications, such as cutaneous squamous cell carcinoma (SCC) (also termed Marjolin ulcer). Herein, we report a case of invasive cutaneous SCC arising from chronic ulcers of a HS patient. A 40-year old Korean male, a current smoker with 20 pack-year history, presented with a history of painful, recurrent, deep-seated abscesses and ulcers on the buttocks since his late teens, thus classified as Hurley stage III. A large purulent ulcer developed on the right buttock several months ago. Initial treatment was focused on controlling infection and facilitating wound healing. The lesion showed 50% reduction of size in 6 weeks, but also developed foul odor and showed fungating margins. Multiple skin biopsies were consistent with invasive SCC. Magnetic resonance imaging revealed a few enlarged lymph nodes on the right inguinal area, which was confirmed as metastasis on frozen biopsy. Slow Mohs micrographic surgery and radical right inguinal lymph node dissection was done. Incidence rates of SCC arising

from HS have been reported up to 4.6%. To our knowledge, this is the first report of cutaneous SCC arising from HS in Korea. Our case emphasizes that the diagnosis of cutaneous SCC in HS should not be delayed, and early surgical intervention is crucial for better outcomes. (**Ann Dermatol 33(1) 68~72, 2021**)

-Keywords-

Carcinoma, squamous cell, Hidradenitis suppurativa, Mohs surgery

INTRODUCTION

Hidradenitis suppurativa (HS) is a chronic, painful, recurrent, and debilitating inflammatory skin disorder of the folliculopilosebaceous units which presents with painful and deep-seated nodules, sinus tracts, and abscesses mainly in intertriginous axillary, groin, perineal, and genital skin¹. Cutaneous squamous cell carcinoma (SCC) may arise in such chronic inflammatory processes, and since first reported in 1958, only about 100 cases of cutaneous SCC arising from chronic HS have been identified. Most cases are treated with wide local excision², which may decrease recurrence rates but also may increase wound size and complications. Slow Mohs micrographic surgery (MMS) is a modified version of conventional MMS, which uses paraffin-embedded sections instead of frozen sections for margin examination. Herein, we report a rare case of cutaneous SCC arising from chronic HS successfully removed with slow MMS.

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CASE REPORT

A 40-year-old male presented with a large, purulent ulcer on his right buttock for several months, accompanied by multiple deep-seated abscesses and sinus tracts on both buttocks (Fig. 1A). He had a 20-year history of HS which started in his late teens. The patient was a current smoker, and reported smoking one-pack of cigarettes per day since his age of 20 years. Initial treatment was focused on systemic antibiotics and antiseptic dressings to reduce burden of infection and facilitate wound healing. The lesion showed some reduction in size after 6 weeks, but also developed foul odor and fungating margins (Fig. 1B). Skin biopsy of the fungating margin showed downward dermal infiltration of irregular masses consisting of atypical epidermal cells and some mitoses (Fig. 2). Although pseudoepitheliomatous hyperplasia (PEH) may be misinterpreted as SCC

in that it shows irregular epidermal hyperplasia and downward infiltrative pattern, SCC can be distinguished from PEH by significant atypical epithelial cells mitoses. Thus in our case, PEH was ruled out and the skin biopsy was consistent with moderately differentiated invasive cutaneous SCC. Multiple additional scout biopsies all showed the same features of cutaneous SCC, even in papules distant from the ulcer (Fig. 1C). Magnetic resonance imaging showed a large infiltrating mass on the skin covering the right buttock and a few enlarged lymph nodes on the right inguinal area with increased signal density (Fig. 3). Chest X-rays and abdominal computed tomography scan showed no sign of distant metastasis.

Due to broadness and infiltrative character, slow MMS was performed with sentinel lymph node biopsy, which proved to be positive for metastasis on frozen pathology. Radical inguinal lymph node dissection of the right side

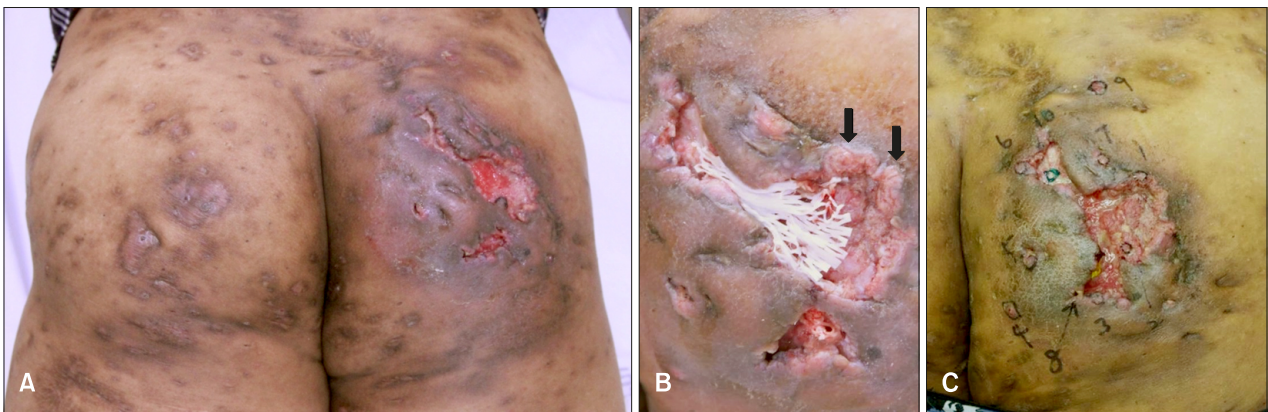


Fig. 1. (A) Clinical appearance of a long-standing ulcer on the right buttock that would not heal for several months. (B) The lesion showed fungating margins (arrows) despite several weeks of conservative treatment. (C) Multiple scout biopsies all showed presence of squamous cell carcinoma, even in papules distant from the ulcer.

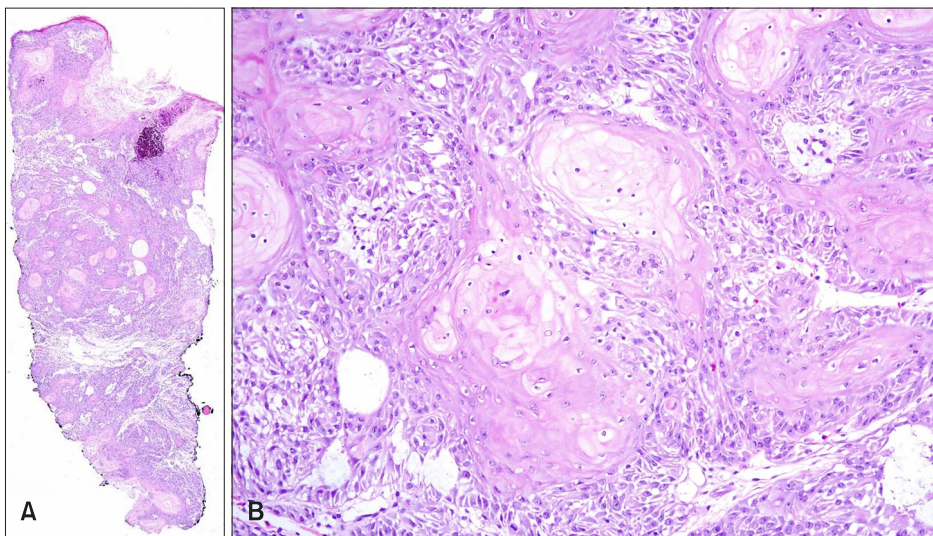


Fig. 2. (A) A biopsy specimen showing downward dermal infiltration of irregular masses (H&E, $\times 40$). (B) Dermal infiltration consisting of atypical epidermal cells and some mitoses, consistent with moderately differentiated invasive cutaneous squamous cell carcinoma (H&E, $\times 200$).

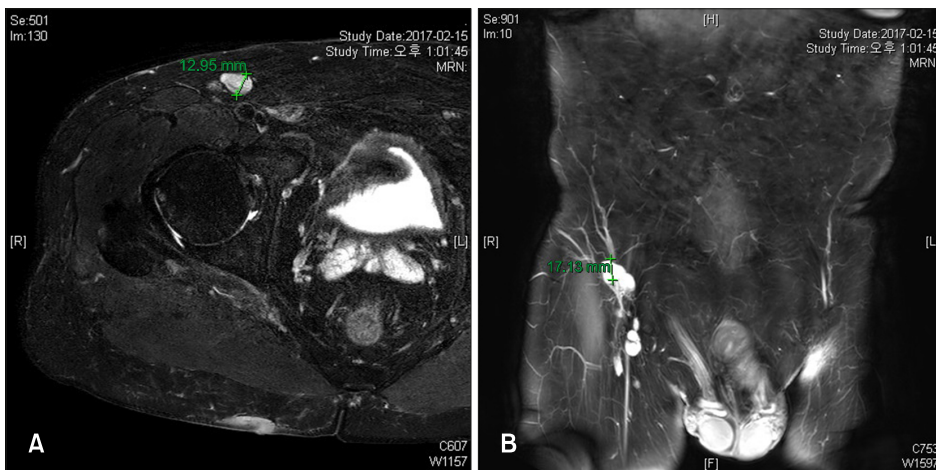


Fig. 3. (A, B) A few significantly enlarged right inguinal lymph nodes (short diameter larger than 1 cm) shown in magnetic resonance imaging, suggestive of regional metastasis.

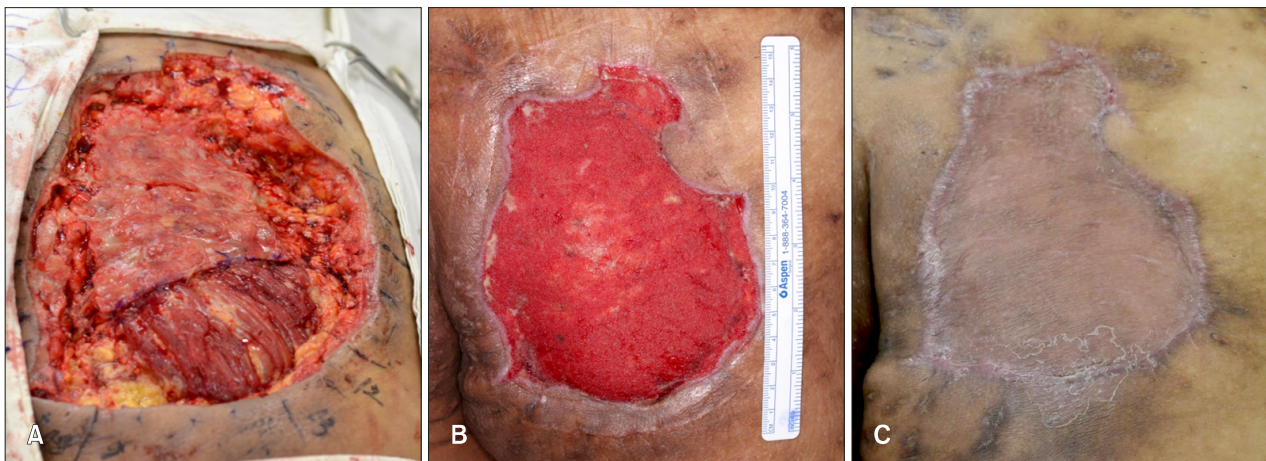


Fig. 4. (A) Defect after 2 stages of slow Mohs micrographic surgery with part of the gluteus maximus muscle removed due to deep infiltration of the tumor. (B) Defect after 2 weeks of negative pressure wound therapy. (C) Defect has healed up well 6 weeks after graft placement.

was done additionally. The pathology of 1st stage of slow MMS revealed presence of carcinoma in 3 sections out of 29 lateral marginal sections, and 3 sections out of 38 base marginal sections. For this reason, the 2nd stage was performed, which showed negative of tumor in the all lateral and base marginal sections. Accordingly the tumor was completely removed in 2 stages (Fig. 4A) with the 2nd stage removing a layer of the fascia and muscle from the gluteus maximus muscle leaving a deep, 15 by 12-cm-sized defect. Due to the large size, the defect was first treated with negative pressure wound therapy for 2 weeks in order to decrease the size and depth (Fig. 4B), and then covered with split thickness skin graft from the right posterior thigh. The defect has healed up well 6 weeks after graft placement (Fig. 4C). For further evaluation and treatment, a whole body positron emission tomography-computed tomography scan, chemotherapy and radiation were recom-

mended, but the patient refused any further evaluation or adjuvant treatment. However, the patient showed no sign of recurrence during two-year follow-up period.

DISCUSSION

Cutaneous SCC arising from chronic HS is rarely reported in the literature, with a reported prevalence of 0.5% to 4.6% for cutaneous SCC¹. In contrast to the fact that females are more likely to develop HS than males, there is a 4:1 ratio of male predominance of cutaneous SCC arising in HS. Usually, cutaneous SCC occurs 20 to 30 years after the onset of HS¹ and approximately more than half of these patients die from the associated morbidities. As noted above, prognosis of cutaneous SCC arising in chronic HS is poor, mainly due to the difficulty in clinically differentiating malignant transformations from chronic lesions,

thus leading to delayed diagnosis and advanced staging at the point of diagnosis³, as seen in our patient. Although systematic review of cutaneous SCC arising in chronic HS is lacking due to its rarity, Lavogiez et al.¹ reported that 3 patients out of 13 cases of HS complicated by SCC had local relapse within a few months after wide local excision. Malignant transformation into cutaneous SCC in HS may be explained by impaired Notch signaling, which acts as an epidermal tumor suppressor in cutaneous SCC⁴. It also may induce excessive production of interleukin (IL)-1 β , an important inflammatory mediator in the pathogenesis of HS. There is some evidence that the NLRP3 inflammasome and IL-1 β -induced inflammation promote tumor growth and metastasis⁵, although the exact correlations to cutaneous SCC arising from chronic HS is yet to be explained. In addition, considering the fact that tumor necrosis factor- α (TNF- α) is an important factor in both pathogenesis of HS⁶ and cutaneous SCC⁷, TNF- α could play a role in malignant transformation of HS into SCC. However, further studies are necessary to investigate the specific role of TNF- α in malignant transformation of HS into SCC.

There are no studies comparing the outcomes of MMS and wide local excision for cutaneous SCC arising from chronic HS yet. Wide local excision is considered as a first-line treatment, and some recommend a minimum surgical margin of 2 cm for wide local excision¹. Although implicated in most cases, recurrence and mortality rates are high, even for curative surgery with negative margins³. Also, since most cases occur in perineal, perianal, or gluteal areas³, the surgical outcome of wide local excision may be severely debilitating for the patient, and indefinite margins of the lesion make decision of surgical margins difficult. Therefore, MMS should be considered in the treatment of cutaneous SCC arising from chronic HS, although further comparative studies are needed. MMS has the potential to not only reduce postoperative defect size but also decrease long-term recurrence rates compared to wide local excision, as demonstrated in multiple skin cancers such as basal cell carcinoma, dermatofibrosarcoma protuberans, and extramammary Paget's disease. A modified version of MMS was performed in this case, because most of the tumor tissue was attached to the subcutaneous fat tissue, which may lead to false negative results with frozen pathology. Also, due to an extensive amount of specimens, the frozen sections would have taken considerable time.

In conclusion, we report the first case of cutaneous SCC arising from chronic HS in Korea successfully removed by slow MMS. Cutaneous malignancy should always be suspected and ruled out in long-standing atypical ulcerative lesions of HS, especially for high-risk patients. Also, slow

MMS may be an effective surgical method in decreasing postoperative defect size and preventing recurrence.

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CONFLICTS OF INTEREST

The authors have nothing to disclose.

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Research data are not shared.

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