

Complex Reconstruction of a Pelvic Marjolin's Ulcer Arising from Hidradenitis Suppurativa

Lauren E. Buchanan, MD*
Chris A. Campbell, MD, FACS†

Summary: The chronic inflammation of hidradenitis suppurativa can cause painful nodules, draining abscesses, sinus tracts, and fibrous scars. This long-term cutaneous inflammation in rare circumstances can lead to malignant transformation producing an aggressive cutaneous malignancy referred to as a Marjolin's ulcer. Particularly when a Marjolin's ulcer involves the sacral region, resection and reconstruction can be challenging. We present the case of a patient with a recurrent Marjolin's ulcer originating from a hidradenitis wound bed overlying and involving the sacrum. Previous radiation, large defect size, and sacral and perianal involvement necessitated the use of a multiflap approach. An extended transpelvic vertical rectus abdominis myocutaneous flap, bilateral gluteal advancement flaps, and a delayed transverse back flap were used to reconstruct the defect after abdominoperineal resection and nerve-sparing partial sacrectomy. Flap choice was derived by dividing the defect into anatomic subunits and considering intrapelvic defect volume, creating a systematic approach that led to successful reconstruction and functional restoration. (*Plast Reconstr Surg Glob Open* 2019;7:e2054; doi: 10.1097/GOX.0000000000002054; Published online 23 May 2019.)

Long-term inflammation from the nodules and draining sinuses of Hidradenitis suppurativa (HS) can lead to a Marjolin's ulcer—a rare, aggressive cutaneous malignancy.^{1–3} Arising from 3% of HS cases,⁴ Marjolin's ulcers are aggressive cutaneous tumors most commonly of the squamous cell carcinoma (SCC) type.⁵

The most effective treatment for HS-derived Marjolin's ulcers is surgical excision with adjuvant radiotherapy as indicated based on tumor staging.^{6–8} We present a rare case of Marjolin's ulcer arising from long-standing HS overlying the sacrum that recurred despite resection and radiotherapy. Surgical treatment for this patient required partial sacrectomy, abdominoperineal resection (APR), and a combination of transpelvic and local flaps for perineal reconstruction and functional restoration.

CASE REPORT

We present the case of a 47-year-old man with a history of SCC-type Marjolin's ulcer related to a 29-year history of HS of the gluteal and sacral regions. Before referral,

the tumor was excised with coccygectomy and right posterior thigh flap closure. Three months postresection, the patient's recurrence was treated with radiation with subsequent remission by report.

The patient was subsequently referred to our clinic 2 years later with loss of fecal continence and an 8×6-cm ulcerated wound overlying the sacrum (Fig. 1A).

Local biopsies confirmed SCC recurrence. Preoperative staging computed tomography (CT) revealed a large soft tissue ulcerative lesion with involvement of the anus but without signs of lymphadenopathy or metastasis. Magnetic resonance imaging (MRI) demonstrated S5 vertebral involvement (Fig. 1B).

A multidisciplinary team, including orthopedics and colorectal surgery, developed the surgical plan to resect the Marjolin's ulcer through partial nerve-sparing sacrectomy of S4–5, and APR due to perianal involvement, proximity to the rectum, and incontinence.

The planned pelvic reconstruction was designed through an anatomic subunit approach as described previously by the senior author.⁹ The perineal raphe, perianal skin, pelvic dead space, and midline gluteal areas were to be reconstructed with an extended vertical rectus abdominis myocutaneous flap (eVRAM), the paramedian gluteal defect with remnant bilateral gluteal advancement flaps, and lumbosacral resurfacing with a delayed transverse back flap (Fig. 2).

Two weeks before the planned en bloc resection, an excisional biopsy of the ulcerated mass was performed to confirm clear circumferential cutaneous margins.

From the *Department of Plastic Surgery, University of Virginia School of Medicine, Charlottesville, Va. and †Department of Plastic Surgery, University of Virginia Health System, Charlottesville, Va. Received for publication April 23, 2018; accepted October 12, 2018.

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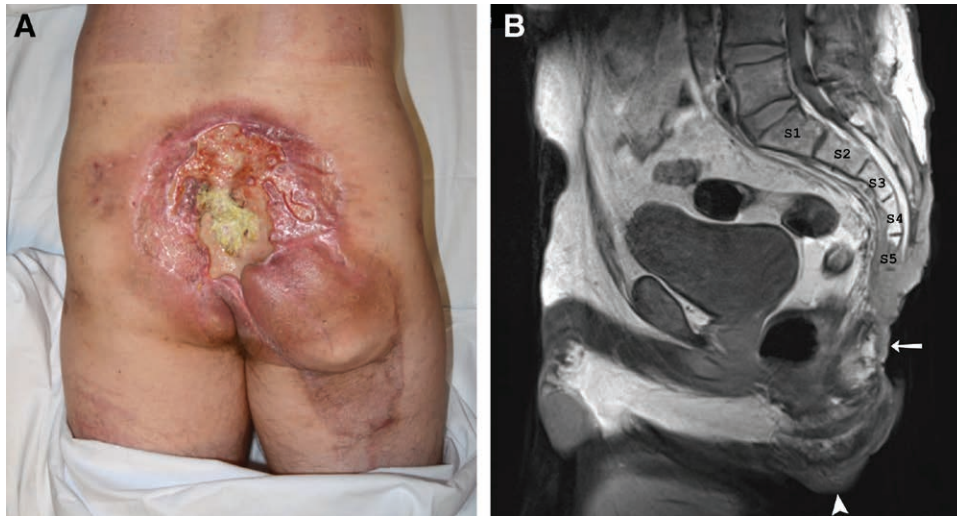


Fig. 1. Pre-operative examination and imaging. A, Preoperative photograph of patient with long-standing hidradenitis suppurativa with biopsy-proven recurrent Marjolin's ulcer of the squamous cell carcinoma (SCC) variety. The patient has had prior resection with right posterior thigh flap closure and radiotherapy. B, Sagittal MRI showing ulcer base (white arrow), tumor invasion of S5 vertebral level, and absence of coccyx from prior resection. Enhancement and thickening of perianal tissues is also evident (arrowhead).

This operation served as an opportunity to delay the blood supply of the transverse back flap. By elevating the flap, the right paraspinous perforator angiosome was able to extend to the tip of the flap on the patient's left posterior back.

The planned eVRAM flap was designed by extending the traditional VRAM skin paddle along the costal margin to the midaxillary line as previously described.¹⁰ Paddle length was confirmed radiographically by measuring the path of flap passage from the inferior epigastric pedicle over the dome of the bladder, to the planned S4–5 spinal level of resection.

Two weeks later, en bloc partial sacrectomy and APR were performed creating an intrapelvic volume defect of 1723.5 cm³. According to the prior sacrectomy case series, this is a medium to large volume defect requiring VRAM for obliteration of pelvic dead space.^{11,12} In contrast to the conditions treated in these series, the large HS cutaneous defect not reflected in the volume assessment required the addition of bilateral gluteal myocutaneous advancement flaps for perineal resurfacing in addition to the transpelvic eVRAM.

The lumbosacral subunit was reconstructed with the delayed transverse back flap which provided significant surface area and ideal thickness to cover the remnant sacrum.¹³

Abdominal wall donor site reconstruction was aided by rectus fascial-sparing harvest of the eVRAM flap and right unilateral component separation as previously described.^{14,15}

Six months postreconstruction, both the donor and recipient sites were well healed (Fig. 2B and C). The patient maintained the ability to urinate spontaneously and was able to sit without discomfort or pressure sore development. His abdominal wall exhibited an asymptomatic right-sided abdominal bulge when bearing down but oth-

erwise his contour was appropriate allowing for the completion of activities of daily living without an abdominal binder. He was most recently examined after requiring a laparotomy and small bowel resection due to obstruction 3 years postoperatively without evidence of hernia or other disability.

This patient's clinical presentation and management demonstrate the vicious, recurrent nature of Marjolin's ulcer¹⁶ requiring multidisciplinary staging and planning. The significant pelvic wound that can result from en bloc tumor resection in the setting of prior radiation can be reconstructed by dividing the pelvic defect into subunits.⁹ In this case, successful reconstruction required a combination of transpelvic and local flaps used to reconstruct a complex pelvic defect and restore form and function.

DISCUSSION

In approximately 3% of cases of HS, chronic lesions result in malignant transformation into a Marjolin's ulcer, most commonly SCC type.^{4,5} Even though malignant transformation normally takes many years to develop, a Marjolin's ulcer once formed behaves aggressively with local destruction and metastasis.¹⁷ A higher metastasis rate at presentation (27.5%) and recurrence rate (58%) are seen in SCC-type Marjolin's ulcer than in all other forms of cutaneous cancer.^{5,6}

The mainstay of treatment of Marjolin's ulcer is surgical clearance.⁶ Because positive histological margins correlate with higher recurrence and metastasis rates, obtaining negative margins is critical and may create large defects that would otherwise compromise cosmesis and function without proper planning.^{7,8}

In our patient, prior radiation, sacrectomy and APR created a large volume dead space in a difficult wound environment that necessitated the use of bulky, well-perfused tissue. Published algorithms for dead space obliteration

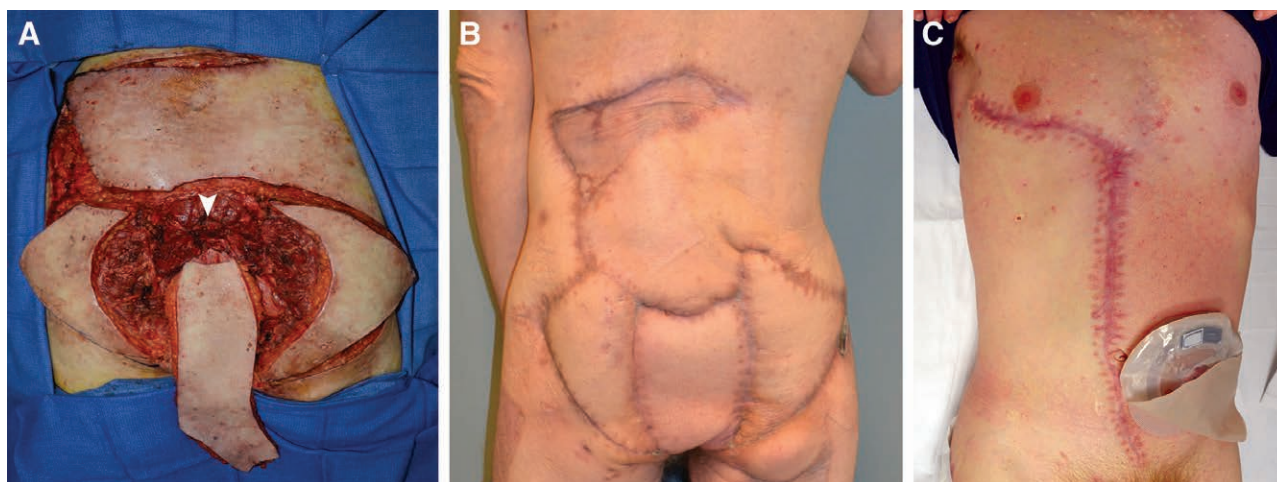


Fig. 2. Intra-operative and post-operative photographs. A, Defect after partial sacrectomy (levels S4 and S5) and APR. Previously delayed transverse back flap has been re-elevated and bilateral gluteal remnant V-Y advancement flaps have also been elevated. Extended vertical rectus myocutaneous (eVRAM) is visible emanating from under S3 (white arrowhead), having passed through its transpelvic course over the bladder and under the remaining sacrum. B, Six-month follow-up after partial sacrectomy and abdominoperineal resection (APR), with eVRAM flap, bilateral gluteus myocutaneous advancement flaps and transverse back flap reconstruction, posterior view. C, Abdominal donor site view at 6 months.

tion after sacrectomy begin with volume analysis to decide between VRAM for larger volume defects or gluteal flaps for smaller defects.^{11,12} The combined sacral and APR that this patient required created a large intrapelvic volume defect that would require VRAM closure.

The resection of ulcerated midline posterior and lateral buttock skin regions required large surface area skin coverage that can be divided into perineal and buttock subunits.⁹ Classically, midline defects have been reconstructed with the VRAM, whereas defects consisting of midline to lateral skin resections were reconstructed with gluteus advancement. An extended VRAM flap was chosen to resurface the midline pelvic defect due to its larger volume and greater skin paddle surface area. Remnant gluteal advancement flaps were used for the paramedian perineal resurfacing as the medial half of the traditional gluteus V-Y skin paddle had been resected with the tumor.

The lumbosacral region is outside of the classic perineal anatomic unit necessitating the use of the delayed transverse back flap which provided significant surface area and ideal thickness to cover the remnant sacrum.¹³ In our case, the transpelvic, presacral course of the eVRAM directed the skin paddle inferiorly to cover the midline buttock and perianal defect, preventing it from covering the proximal sacrum. The transverse back flap was appropriate for coverage of this subunit.

Rectus fascial-sparing harvest and component separation techniques were performed to facilitate donor site closure without the need for surgical mesh. These techniques have been associated with lower rates of postoperative seroma, skin dehiscence, and hernia after VRAM harvest consistent with our findings of an asymptomatic abdomen for our patient 3 years after reconstruction.^{14,15}

As this treatment plan demonstrates, the extensive nature of this defect required a combination of transpelvic and local flaps that can be applied and modified in similar

cases depending on pelvic and lumbosacral anatomic unit involvement.

SUMMARY

The chronic inflammation of hidradenitis suppurativa can cause painful nodules, draining abscesses, sinus tracts, and fibrous scars. This long-term cutaneous inflammation in rare circumstances can lead to malignant transformation producing an aggressive cutaneous malignancy referred to as a Marjolin's ulcer. Particularly when a Marjolin's ulcer involves the sacral region, resection and reconstruction can be challenging. We present the case of a patient with a recurrent Marjolin's ulcer originating from a hidradenitis wound bed overlying and involving the sacrum. Previous radiation, large defect size, and sacral and perianal involvement necessitated the use of a multiflap approach. An extended transpelvic vertical rectus abdominis myocutaneous flap, bilateral gluteal advancement flaps, and a delayed transverse back flap were used to reconstruct the defect after abdominoperineal resection and nerve-sparing partial sacrectomy. Flap choice was derived by dividing the defect into anatomic subunits and considering intrapelvic defect volume, creating a systematic approach that led to successful reconstruction and functional restoration.

Chris A. Campbell, MD, FACS

Department of Plastic Surgery
University of Virginia Health System
Charlottesville, VA 22908

E-mail: cac5rb@hscmail.mcc.virginia.edu

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