

## CASE REPORT

# Advanced Hidradenitis Suppurativa Presenting with Bilateral Vulvar Masses

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**Hidradenitis suppurativa is a chronic suppurative inflammatory disease of the apocrine sweat glands. It is more common in women and involves the genital region in half of all affected patients. An advanced case of hidradenitis suppurativa with bilateral vulvar masses is presented. Possible etiologies of the disease are discussed, as well as strategies for definitive therapy. Because the disease frequently affects the female external genitalia and therapy can require extensive surgical resection and reconstruction, the services of a gynecologic oncologist may be required. Adequate therapy based on an understanding of the pathophysiology of the disease can yield significant relief of symptoms.** © 1996 Academic Press, Inc.

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### INTRODUCTION

Hidradenitis suppurativa is a chronic suppurative inflammatory disease involving the apocrine sweat glands in the axilla, genital area, breast, and other sites. Because the disease can produce malignant-appearing lesions of the vulva and require extensive surgical therapy, the gynecologic oncologist may be called upon to make a diagnosis and render treatment of this benign condition. We present a case where the patient presented with lesions of the genital region accompanied by large bilateral vulvar masses. She was initially referred to the division of gynecologic oncology to rule out a vulvar malignancy. Treatment required resection of the vulva with adjacent areas of involvement, followed by reconstructive surgery. The pathophysiology of the disease as well as effective therapies are discussed.

### CASE REPORT

A 41-year-old white female was referred to the gynecologic oncology clinic because of large vulvar masses believed to represent a vulvar malignancy. The patient had noted several boils in both axillae that coalesced and formed multiple draining sinus tracts 10 years prior to her present

illness. The axillary process resolved spontaneously, but the patient subsequently noted a similar condition on her vulva. She refused to seek further medical attention for this condition and over the next 10 years the inflammatory disease progressed. The vulvar masses had produced considerable foul-smelling drainage and frequent bleeding for the preceding 2 years. At the time of presentation, the masses were very painful. The patient also admitted to malaise, lethargy, night sweats, and anorexia with a 10-pound weight loss over the past month. The remainder of the patient's history was significant for a 23 pack-year history of tobacco use and a 5- to 10-year history of alcohol abuse.

On physical examination, the patient was unkempt and appeared nervous. She was wearing an adult diaper which served as a sling to support bilateral vulvar masses. The vulva was completely distorted. Two pedunculated masses, each with a stalk measuring 4 to 5 cm in length and 2 to 3 cm in thickness, protruded from the left and right intralabial folds (Fig. 1). Each mass measured approximately 20 cm in total length and 10 cm in diameter. The masses were multinodular with papillary projections. The overlying skin was intact but erythematous. There were numerous fistulous tracts which exuded a foul-smelling discharge. The skin over the rest of the vulva showed extensive thickening, scarring, erythema, and numerous fistulous tracts. These changes encompassed the entire skin surface from 5 cm above the pubic symphysis, posteriorly across the perineum and buttocks to a level just above the iliac crests, and laterally to encompass the labia majora, groins, and medial thighs. Speculum examination could not be performed, but the vagina was palpably normal.

Multiple biopsies revealed chronic inflammation with ulceration, giant cells, and scarring. There was no evidence of carcinoma. Based on the patient's clinical presentation and histology, a diagnosis of severe hidradenitis suppurativa was made. The patient was sent home from the clinic on amoxi-



**FIG. 1.** Appearance of the vulva at initial presentation. The two masses were supported by an adult diaper.

cillin/clavulinate 500 mg three times per day and was instructed to apply sulfamylon cream to all affected areas once a day, to be subsequently rinsed off by a sitz bath.

The patient returned to the hospital for admission 4 weeks later and her condition was noted to have markedly improved. The foul odor was absent, and the masses appeared clean and dry with little of the previously seen exudate. The morning after admission, she was taken to the operating room where a superficial but extensive vulvectomy was performed. The dissection included the entire vulva, perineum, perianal area, mons pubis, groins and medial thighs, and the buttocks as far posteriorly as the operating room table would permit.

Postoperatively, she was kept at bed rest except for sitz baths twice daily and after each bowel movement. On Postoperative Day 7, the patient was returned to the operating room for resurfacing of her wound. Her abdominal pannus was undermined and mobilized, and the flap was advanced over the anterior defect. Split thickness skin grafts were harvested from both medial thighs. The skin was then meshed and inset into the perineal defect to cover an area of approximately 700 cm<sup>2</sup> (Fig. 2). The posterior aspect of the perineum remained uncovered. A sterile dressing was applied and the hips were immobilized using a plaster cast.

Postoperatively, the grafts and abdominal flap appeared to heal well. Seven days after her initial grafting, the patient was returned to the operating room for resection of two areas of residual disease over the upper buttocks, measuring 6 × 9 and 4 × 4 cm, that were not previously accessible. A 0.012-in. split-thickness skin graft was obtained from the left posterolateral thigh, meshed, and grafted over the posterior perineum and buttocks. Although all grafts took well, the previously advanced abdominal flap developed progressive necrosis. Therefore the patient was taken back to the operating room 14 days after the initial grafting procedure. The necrotic areas of the abdominal flap were debrided. A 240-

cm<sup>2</sup> defect was covered with 0.012-in. split-thickness skin grafts harvested from the left thigh. The patient was discharged from the hospital in good condition 8 days after her last procedure and 30 days after her initial surgery.

The patient was seen in frequent follow-up for the next 5 months. She was instructed to scrub the area thoroughly with hexachlorophene solution. A 5 × 15-mm area of recurrence was noted 2 months after discharge, but resolved with debridement and hexachlorophene scrubs. After 48 months, she remains free of recurrence.

## DISCUSSION

Hidradenitis suppurativa, first described by Velpeau in 1839 [1], is a chronic, recurrent suppurative inflammatory disease of the apocrine sweat glands. The early stages of the disease are characterized by painful subcutaneous nodules in areas containing apocrine sweat glands. Chronic infection results in fibrosis, hypertrophic scarring, and multiple sinuses with malodorous drainage. The disease is much more common in women than in men, with one series reporting a 13:5 ratio [2]. An increased incidence is observed among blacks, possibly because of a greater density of apocrine glands [3]. The most common sites affected are the axilla (61%) and groin (48%), followed by the perianal area (22%), breast (13%), and other sites, including the scalp (13%) [4]. Multiple sites are involved in 65% of patients [4]. The disease primarily affects young adults. The mean age at onset is 25 years and it almost never occurs before puberty [2].

The differential diagnosis generally includes furunculosis, pyoderma, granuloma inguinale, acinitis, lymphadenitis, actinomycosis, scrofula, erysipelas, lymphogranuloma venereum, pilonidal disease, inflammatory bowel disease, and immunodeficiency syndromes [5]. With the pedunculated



**FIG. 2.** Appearance of the wound after the first skin grafting procedure. An abdominal flap has been advanced over the anterior portion of the wound, and the groin areas were closed primarily. Split thickness skin grafts from the medial thighs have been used to cover the remainder of the wound.

masses seen in this patient, verrucous carcinoma and extensive condyloma accuminata were also considered. As with any chronic irritative process, a concurrent squamous cell carcinoma must be excluded. Although most reported cases of squamous cell carcinoma developed in patients with long-standing disease, Zachary reported a patient who developed cancer just 3 years following the onset of symptoms of hidradenitis suppurativa [6].

The disease generally begins with the plugging of an apocrine duct. This leads to obstruction and dilation of the apocrine gland, followed by local secondary infection. Rupture of the infected gland results in the infection of adjacent glands and progression of the disease. The most common organisms are staphylococci, but the process can include multiple gram-negative and anaerobic bacteria, especially in the perineal area [5]. It is unclear what makes certain individuals especially susceptible to this process. The disease can sometimes be induced experimentally by depilation and plugging of the apocrine glands with keratinous material [7]. While chemical depilatories or deodorants have been implicated in the past, a retrospective case-control study failed to find such an association [8]. There may be a genetic form of the disease, transmitted in an autosomal dominant pattern [9]. Because the disease is almost never seen before puberty, endocrine abnormality, especially increased androgenicity, may play a role. Harrison *et al.* [2] found that 57% of women experienced a premenstrual flare of their disease, and were more than twice as likely as a control population to be overweight (defined as body mass index greater than 25 kg/m<sup>2</sup>) and to have increased androgen levels. Similar findings have been reported by Mortimer [10]. Camisa *et al.* reported a patient successfully treated in part with leuprolide acetate [11]. The patient initially responded to conservative therapy with clindamycin, isotretinoin, and dexamethasone. However, she recurred multiple times and seemed to be dependent on high-dose steroids for continued remission. The addition of leuprolide permitted the discontinuation of all other medications except low-dose dexamethasone while the patient had continued improvement in her clinical condition. Camisa believed that the patient's response was due, at least in part, to androgen suppression from chronic leuprolide administration.

Conservative surgical therapy, consisting of incision and drainage, was first described by Aristide Verneuil in 1854 [12], and for many years the clinical syndrome was known as Verneuil's disease. Since his initial report of treatment of hidradenitis suppurativa, numerous medical therapies have been tried. Historically zinc peroxide, sulfur baths, ultraviolet radiation, injection of whole milk, immunotherapy with typhoid vaccine or staphage lysate, and local irradiation have all been used in the management of hidradenitis suppurativa [5, 13–16]. More modern conservative treatment generally consists of good hygiene, broad spectrum antibiotics, warm

compresses or sitz baths, and local incision and drainage of fluctuant nodules.

In advanced, recurrent cases of hidradenitis suppurativa, the only appropriate therapy is wide local excision of all affected areas, including adjacent areas of skin containing apocrine glands. There are several options for skin closure. If the defect is small enough and the adjacent skin can be well mobilized, direct reapproximation of skin edges is sometimes possible. However, many surgeons are hesitant to undertake primary closure over an infected site. Pollock [17] and Tache [18] have reported good results with primary closure, but Watson [19] reported a 54% recurrence rate using this technique. Larger defects may be immediately closed using skin grafts or advancement of local flaps, but this may have the same disadvantages as immediate primary closure. The patient described in this report underwent delayed closure with local flaps and skin grafts, allowing antibiotic therapy and mechanical debridement during the interval between the primary surgery and delayed closure. Other surgeons have reported favorable experience with healing by second intention [6, 20]. Morgan [21] reported a series of 10 patients with bilateral axillary involvement treated with excision and split-thickness skin grafting on one side, while the contralateral side was allowed to heal by second intention. Although the grafted sites healed faster, Morgan preferred closure by second intention because the cosmetic results and range of motion were just as good, there was no donor site, and no postoperative period of immobilization [19].

We have chosen to present this case because it represents an advanced stage of a nonmalignant vulvar disease which required the services of a gynecologic oncologist. The combination of antibiotic therapy, surgical resection, reconstruction, and delayed closure resulted in a satisfactory outcome.

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