Presentation and surgical management of hidradenitis suppurativa of the breast during pregnancy: A case report

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ABSTRACT
INTRODUCTION: Hidradenitis suppurativa (HS) is a chronic inflammatory disease of the skin and soft tissues most commonly affecting the axilla and groin. It presents as recurrent inflamed lesions, abscesses, draining sinus tracts, and scars. Treatment focuses on preventing progression of disease and managing symptoms. There are limited guidelines discussing the treatment of HS during pregnancy as well as the surgical treatment of HS of the breast in young women who desire future lactation.

PRESENTATION OF CASE: We present a case of a thirty-six-year-old female with HS of the breast during pregnancy. Her condition was recalcitrant to steroid injections, TNF blocker, antibiotics and incision and drainage. She developed numerous active, infected large interconnected lesions in her right breast. The decision was made to perform total excision of the infected area in the right breast. She underwent primary closure of the wound one week after excision and was able to successfully breast feed after this treatment.

DISCUSSION: Treatment of HS includes both medical and surgical modalities and varies based on the severity of disease. In this patient, definitive but conservative pre-partum treatment was necessary to avoid milk fistula or chronic abscess formation, as the patient’s desire was successful postpartum lactation.

CONCLUSION: This report describes the successful surgical management of a pregnant woman with severe HS of the breast. This represents an alternative management method in an extreme case of HS, where aggressive medical management was contraindicated.

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1. Introduction

Hidradenitis suppurativa (HS) has classically been considered a chronic acniform infection of the apocrine glands; however, more recent literature describes it as a chronic follicular occlusive disease [1–4]. HS most commonly affects the axilla and groin but can also involve the inframammary cleft, areola, perineum, gluteal folds, perianal region and inframamillary skin folds [1,2,5,6]. The pathogenesis has been poorly understood and has changed throughout the years. Historically, HS was thought to result from entrapped apocrine secretions, allowing bacteria to proliferate, creating inflammation and infection [1,2]. More recent theories propose a mechanism of follicular occlusion caused by follicular epithelial hyperplasia [3,4]. Regardless of the proposed pathogenesis, the cause of HS has always been multifactorial. Predisposing factors include genetics, environmental factors, obesity, smoking, and hormones [3,7]. The presentation depends on the chronicity of the disease and includes recurrent tender erythematous lesions, abscesses, draining sinus tracts, double-ended comedones, and dermal contractures [2]. The lesions can open and spontaneously drain clear to purulent material and eventually heal with or without surgical intervention [5,6]. Remission may last from weeks to years but some patients may be continuously affected without remission. Patients with continuous inflammation may present with fibrotic transformation of the soft tissues. The differential diagnosis includes pilonidal cyst, lymphogranuloma venereum, and granuloma inguinale. Complications include restricted motility from chronic scarring and contracture in the axillae or groin, lymphedema from obstruction of the lymphatics and squamous cell carcinoma [8]. Diagnosis is based on history, location, and the classic clinical features. Treatment is difficult and the approach depends on the presentation and severity of disease. Described treatments have included lifestyle modification, topical and oral antibiotics, intradermal and systemic steroids, antianogenrels, TNF-alpha inhibitors, and surgery. All of these management strategies focus on minimizing progression of disease, managing symptoms, and restoring quality of life.

Abbreviation: HS, Hidradenitis suppurativa.

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HS is commonly seen in post-pubertal females with a female to male ratio of 3:1 [7]. The post-pubertal and female predominance has been attributed to hormonal stimulation of the affected glands; however, the role of hormones in the pathogenesis of HS has not been validated [7,9,10]. Most women continue to experience symptoms during pregnancy which poses a challenge to treatment [11]. Many medications are contraindicated in pregnancy and general anesthesia poses a risk to the fetus during surgery. The objective of this report is to describe the presentation and successful management of a severe case of HS of the breast in a pregnant African American female. This work has been reported in line with the SCARE criteria [12].

2. Presentation of case

A thirty-six-year-old African American woman, gravida 1, par- turition 0, presented to our community-based, university-affiliated teaching hospital at 26 weeks gestation with worsening HS of her right breast. She had a 30-year history of HS involving bilateral breasts, axillae, and groins. She was treated by a dermatologist with steroid injections and adalimumab for six months prior to her presentation. She was morbidly obese with a BMI of 47.5 and a non-smoker.

On physical exam, the right breast had acute and chronically inflamed draining nodules involving the areola and breast from the 3 o’clock to 8 o’clock position. The left breast and both axillae had healed scars from previous hidradenitis infections.

Following conception of her pregnancy, she discontinued adalimumab, with subsequent worsening of her HS. Over the course of two months, she presented several times to the emergency department; twice with cellulitis and abscess of the right breast for which she was prescribed oral antibiotics and discharged home. At her third presentation, she had fever, tachycardia, leukocytosis, right breast cellulitis and actively draining nodules.

At this presentation, she was admitted for IV antibiotics and a surgical consult. A bedside ultrasound revealed an abscess which was not amenable to fine needle aspiration due to the thickness of the purulent fluid. She was taken to the operating room and underwent incision and drainage of the right breast and placement of a Jackson Pratt drain (Fig. 1). Her wound cultures grew *Streptococcus constellatus*, *Pseudomonas aeruginosa*, and methicillin-susceptible *Staphylococcus aureus*. She was discharged home with six weeks of IV meropenem and piperacillin-tazobactam. Her drain was inadvertently dislodged two weeks after placement and a right breast ultrasound did not show any recurrent fluid collections.

She was seen four weeks after the completion of the IV antibiotics and was noted to have a large central mass of the right breast with multiple scars and erythematous, draining nodules. At this time, she was admitted to the obstetrics service with consultation provided by breast surgery and infectious disease. The differential diagnosis included HS, breast abscess, and inflammatory cancer. The patient refused mammogram with abdominal shielding due to her concern for fetal radiation exposure. MRI of the breast was not an option due to gadolinium crossing the placenta and the potential for subsequent fetal abnormalities. Therefore, she underwent ultrasound of the right breast which showed loculated interconnected abscesses.

Given her unrelenting condition, she was taken to the operating room for a wide local debridement and excision of the affected area (Fig. 2) with continuous fetal monitoring. The breast tissue was submitted to pathology for aerobic, anaerobic, and fungal cultures and to rule out underlying malignancy. She was treated with piperacillin-tazobactam and meropenem and subsequently developed bilateral lower extremity erythema nodosum. Her treatment was then changed to vancomycin, cefazidime, and metronidazole.

Pathology revealed acute on chronic inflammation with areas of organizing abscesses (Figs. 3 and 4) but no evidence of any organism on gram stain or cultures. She was managed with wet to dry dressings for two days post-operatively and then negative-pressure wound therapy for five days. She was taken back to the operating room for delayed primary closure of the wound with nylon vertical mattress sutures. She was seen 14 days post-operatively for suture
Fig. 4. Pustule formation with adjacent cystic follicle filled with keratinous debris (hematoxylin – eosin, ×200).

Fig. 5. Right breast 16 days after primary closure. There are no signs of recurrent or persistent infection. The scars from the interrupted sutures are expected to improve over time.

removal and her wound was healing well without any signs of infection (Fig. 5). After delivery she was able to successfully breast feed her baby, and after a year of follow up, she has had no recurrences in that breast.

3. Discussion

Among the list of chronic inflammatory cutaneous disorders, HS is at the top of the list for adversely affecting quality of life [13]. The patient, a 36-year-old morbidly obese pregnant female, suffered through a 30-year history of recurrent and recalcitrant HS despite multiple varying treatment modalities.

The European S1 guideline recommends medical treatment as monotherapy or combined with radical surgery for the management of widely spread lesions [14]. It could be argued that she should have been treated with antibiotics for a longer duration than six weeks given that she returned with a large right breast abscess four weeks after the completion of the antibiotics. However, there is limited evidence that long term antibiotic therapy cures HS; with relapse often occurring after the cessation of therapy [9].

In this case, the goal was to manage the HS surgically while considering the patient’s wishes for future lactation and potential need for reconstruction depending on the cosmetic outcome. The antibiotics administered during her second admission did reduce the inflammation within the areolar region allowing for preservation of 85% of the nipple-areolar complex at the time of debridement. If there had not been any improvement, the nipple and areola would have been excised with the local debridement. Other important considerations are in counseling the patient about possible difficulties with lactation and milk fistula, including the potential for inability to breast feed.

As an additional precaution in this case, consultation was made to the anesthesia team for a bedside assessment and detailed discussion of the risks of surgery to both mother and fetus. Continuous fetal heart tone monitoring was provided by the obstetrics department to monitor for fetal changes and preterm labor during both the wide-local debridement and the delayed primary closure. In the end, both breasts were symmetric as the acute and chronic inflammation had caused the right breast to be 1.5 times larger than the left prior to debridement and excision.

It is noteworthy that there is a lack of evidence regarding any sole treatment for HS. Prevention is the primary goal. Antibiotics may be used to treat acute exacerbations while focal abscesses may only require drainage. Over time, the disease resolves, leaving behind fibrosis. Surgical treatment is not always successful and can be quite extensive. Effective surgical management requires excision of the entire area including all tissue with fibrosis, sinus tracts, and fistulae [15].

4. Conclusion

This report describes the presentation and successful surgical management of a pregnant African American female with an acute HS exacerbation. The patient’s quality of life was improved for the duration of her pregnancy without placing the fetus at unnecessary risk. She delivered her baby and was successful in lactating from bilateral breasts. This case presents an alternative management method in an extreme case of HS, where aggressive medical management was contraindicated.

Conflict of interest

The authors have no conflict of interest to declare.

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Ethical approval

This is not required for a case report. Approval to publish this case report was waived by the institution.

Consent

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Authors contribution

Selyne Samuel, MD: drafted and critically revised article, surgeon involved in diagnosis and management plan.
Abigail Tremelling, MD: critically revised and completed final revisions of article.
Mary Murray, MD: critically revised article, surgeon involved in diagnosis and management plan.
Registration of research studies

We do not need to register this work.
Not applicable.

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References

[1] R.F. Edlich, K.A. Silioway, G.T. Rodeheaver, Epidemiology, pathology, and treatment of axillary hidradenitis suppurativa, J. Emerg. Med. 4 (1986) 369–378.
[2] G.B. Jemec, M. Heidenheim, N.H. Nielsen, Hidradenitis suppurativa – characteristics and consequences, Clin. Exp. Dermatol. 21 (1996) 419–423.
[3] H.H. van der Zee, J.D. Laman, J. Boer, E.F. Prens, Hidradenitis suppurativa: viewpoint on clinical phenotyping, pathogenesis and novel treatments, Exp. Dermatol. 21 (2012) 735–739.
[4] M. von Laffert, V. Stadie, J. Wohlhrab, W.C. Marsch, Hidradenitis suppurativa/acne inversa: bilocated epithelial hyperplasia with very different sequelae, Exp. Dermatol. 164 (2011) 367–371.
[5] H. Deroo, M. Aelbrecht, J. t’Kindt, Hidradenitis suppurativa, Dermatologica 180 (1990) 193–194.
[6] R.L. Attanasio, M.A. Appleton, A.G. Douglas-Jones, The pathogenesis of hidradenitis suppurativa: a closer look at apocrine and aepocrine glands, Br. J. Dermatol. 133 (1995) 254–258.
[7] G.B. Jemec, M. Heidenheim, N.H. Nielsen, The prevalence of hidradenitis suppurativa and its potential precursor lesions, J. Am. Acad. Dermatol. 35 (1996) 191–194.
[8] C. Constantinou, K. Widom, J. Desantis, M. Obmann, Hidradenitis suppurativa complicated by squamous cell carcinoma, Am. Surg. 74 (2008) 1177–1181.
[9] B.J. Harrison, M. Mudge, L.E. Hughes, Recurrence after surgical treatment of hidradenitis suppurativa, Br. Med. J. (Clin. Res. Ed.) 294 (1987) 487–489.
[10] G.B. Jemec, The symptomatology of hidradenitis suppurativa in women, Br. J. Dermatol. 119 (1988) 345–350.
[11] P. Perg, J.G. Zampella, G.A. Okoye, Management of hidradenitis suppurativa in pregnancy, J. Am. Acad. Dermatol. 76 (2017) 979–989.
[12] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohalan, D.P. Orgill, for the SCARE Group, The SCARE Statement: consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[13] F.A. Kerdel, A. Menter, R.G. Michelelli, Hidradenitis suppurativa: update on diagnosis and treatment, Semin. Cutan. Med. Surg. 33 (2014) 547.
[14] C.C. Zouboulis, N. Desai, L. Entestam, R.E. Hunger, D. Ioannides, I. Juhasz, et al., European S1 guideline for the treatment of hidradenitis suppurativa/acne inversa, J. Eur. Acad. Dermatol. Venereol. 29 (2015) 619–644.
[15] J.E. Fischer, D.B. Jones, F.B. Pomposelli, G.R. Upchurch, V.S. Klimberg, S.D. Schwartzberg, et al., Mastery of Surgery, 6th ed., Lippincott Williams & Wilkins, Philadelphia, 2012.

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