Case report

# Successful treatment of stage III hidradenitis suppurativa with botulinum toxin A

Weibin Shi,[1,2] Shannon Schultz,[1,2] Amy Strouse,[1,2] David R Gater[1,2]

[1]Physical Medicine and Rehabilitation, Penn State Health Milton S Hershey Medical Center, Hershey, Pennsylvania, USA
[2]Penn State Hershey Rehabilitation Hospital, Hummelstown, Pennsylvania, USA

**Correspondence to**
Dr Weibin Shi,
wshi@pennstatehealth.psu.edu

Accepted 10 December 2018



**To cite:** Shi W, Schultz S, Strouse A, *et al*. *BMJ Case Rep* 2019;**12**:e226064. doi:10.1136/bcr-2018-226064

## SUMMARY

A 41-year-old obese Native American woman presented with hidradenitis suppurativa (HS) after failing all available treatment options. Her HS was more pronounced in the axillary and groin regions (Hurley stage III) and was a major source of her pain and a barrier for home exercise and aquatic therapy. She received a botulinum toxin A injection every 3 months (four times thus far), which has significantly helped alleviate her pain and curb the progress of her HS by resolving abscesses and healing draining sinuses.

## BACKGROUND

Hidradenitis suppurativa (HS) is a common, chronic, inflammatory skin disorder.

The clinical manifestations vary, ranging from recurrent papules, pustules and a few inflammatory nodules to deep fluctuant abscesses, draining sinuses and severe band-like scar formation. It can be classified by Hurley staging system: stage I: abscess formation, single or multiple, without sinus tracts and cicatrisation; stage II: recurrent abscesses with tract formation and cicatrisation, single or multiple, widely separated lesions; and stage III: diffuse or near-diffuse involvement, or multiple interconnected tracts and abscesses across the entire area.[1] Its associated pain, malodour, drainage and disfigurement contribute to a profound psychosocial impact on these patients' lives. Patient education and psychological support are additional important components of patient management. Currently, there is a lack of consensus on optimal management of HS. The overall treatment goal is to limit its progression in all stages of the disease by preventing new lesions. Treatment modalities include patient self-management (avoidance of skin trauma, smoking cessation and weight management), medical (topical therapy, oral systemic agents and biologic therapies) and other interventions (laser/light interventions, surgery).[1] For recalcitrant stage III HS refractory to all conservative treatments, periodic surgical excision could be the only option. There are a handful of successful HS cases treated with botulinum toxin reported,[2–4] but mostly involving stage I or stage II disease. Here, we report the successful application of botulinum toxin A (BTXA) for Hurley stage III HS.[1]

## CASE PRESENTATION

A 41-year-old obese Native American woman initially presented to our physical medicine and rehabilitation clinic for evaluation and management of her chronic pain. After investigating the sources of her pain, we found the primary causative agent was her uncontrolled hidradenitis suppurativa (HS), which significantly affected both axillary and groin regions (Hurley stage III). She had been seen by multiple renowned experts and failed all available treatments options, including lifestyle modification, clindamycin (both cream and oral), minocycline, doxycycline, acitretin, rifampicin-moxifloxacin-metronidazole and a few surgical procedures, such as incision and drainage of abscesses, excision of scars and reconstruction. Immunosuppressants and biological agents were not recommended due to her positive purified protein derivative (PPD) test. She had a course of prophylactic treatment 5 years ago for tuberculosis. The manifestations of her HS, including the open draining sinuses and recurrent abscesses, with stage III presentation in her axillary and groin areas, had profoundly affected her both physically and mentally. She used to enjoy aquatic therapy for her chronic pain; however, secondary to these wounds, she was excluded from the aquatic therapy programme.

## DIFFERENTIAL DIAGNOSIS

Hyperhidrosis.
Follicular pyodermas.
Intergluteal pilonidal disease.
Granuloma inguinale.

## TREATMENT

She received her first injection of botulinum toxin A (BTXA) on 16 November 2016. The BTXA was reconstituted with normal saline (100 units in 2 mL normal saline) and was injected intradermally in 1-inch linear parallel track about 1 cm apart with 30-gauge 1-inch needle, five units in 0.1 mL each injection, about 20 injections in one area.[5] No local anaesthesia was used. With our line-block technique, the needle might be inserted subdermally at times. For papules or any inflammatory areas, we injected around them and literally blanched the erythematous areas. We injected bilateral axillary and inframammary fold regions as a trial, with 100 units for each location, including the areas with

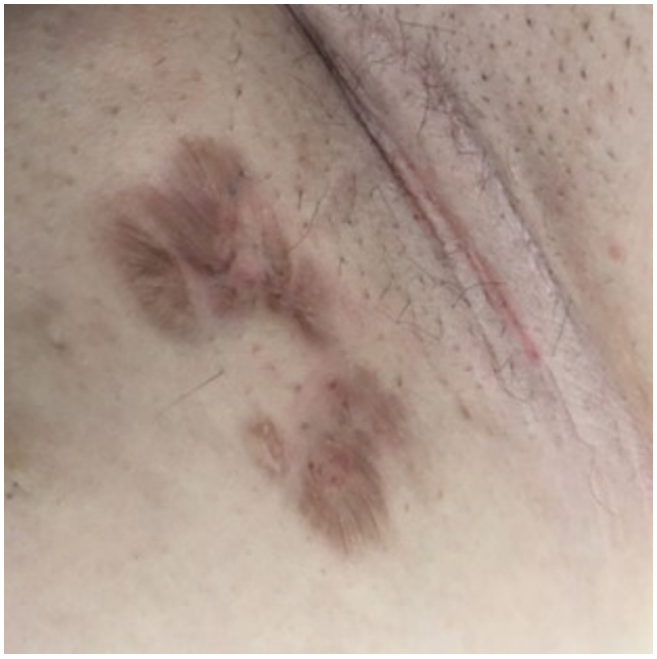

**Figure 1** Healed wound in right axillary area with scar formation after the botulinum toxin A injection.

frank inflammation (400 units total). She had an excellent response on follow-up examination, with a significant reduction in inflammation and drainage with healing of some sinus tracts. She also reported overall at least 50% pain relief. Since the HS in the inframammary region was under good control after the first injection, we decreased the dose of BTXA to 50 units for each location. In addition, we extended the injection sites to her groin and perineal regions with the next two injections. However, her symptoms returned much earlier with the lower dosage. Therefore, starting from the fourth injection, we increased the dosage back to 100 units for each area (bilateral axillary, inframammary and groin) with some additional BTXA into affected glands in the postauricular and torso regions. No other treatments have been received during the BTXA therapy.

## OUTCOME AND FOLLOW-UP

With the dose adjustment, a favourable effect was reproduced successfully. Resolution of inflammation and healing of draining sinuses was seen in all involved areas, including the axillary and groin regions with initial stage III presentation (figure 1). She has been open-wound-free since the fifth injection. Overall, her HS is under good control with periodic BTXA injection.

## DISCUSSION

As the name implies, hidradenitis suppurativa (HS) was previously considered a suppurative disorder primarily involving sweat glands, in particular, apocrine sweat glands. HS is now considered a chronic follicular occlusive disease, even a systemic inflammatory condition, involving the follicular portion of folliculopilosebaceous units (FPSUs).[6] Occlusion of hair follicles occurs through keratinocyte plugging due to its failure of terminal differentiation, with subsequent engorgement with follicular components and apocrine secretions. This results in rupture of the follicle, dissemination of macro follicular contents and inflammation. Over time, the initial inflammatory response may evolve into a chronic foreign body-type granulomatous inflammation. The common sites of involvement for HS are the intertriginous skin areas, including the axillary, groin, perianal, perineal and inframammary regions.

Botulinum toxins block the release of acetylcholine and a number of other neurotransmitters from presynaptic vesicles by deactivating SNARE proteins. It has been used for hyperhidrosis by blocking the postganglionic sympathetic cholinergic nerve fibres to the sweat glands. In 2004, the US Food and Drug Administration approved onabotulinum toxin A for the treatment of severe primary axillary hyperhidrosis in patients unable to obtain relief using antiperspirants.

The exact mechanism by which botulinum toxin affects the disease process in HS remains unclear. Although current consensus on HS pathology is chronic follicular occlusive disorder, the fact that HS mainly affects apocrine-gland-bearing skin still implies apocrine glands play an important role. First, unlike the solitary eccrine glands, apocrine glands are associated with FPSU and discharge in the canal of hair follicles. This likely contributes to the pressurisation and subsequent rupture of the follicle. The spillage of its contents, including corneocytes, bacteria, sebum products and hair, through the surrounding dermis, elicits a chemotactic response and resultant inflammatory cell infiltrate. Reducing apocrine secretions will likely decompress the follicle and prevent it from rupture. Second, apocrine glands play a pivotal role in the inflammatory process of HS. In addition to pressurising the follicle, the entrapped secretion in the apocrine-gland-bearing skin regions provides ideal culture medium for skin flora. Blocking apocrine secretion would presumably reduce the population of skin flora and hence the inflammation. It has been hypothesised that these inflammatory events in patients with HS are secondary to an underlying aberrant inflammatory state instead of the primary cause of the disease.[7] In this case, it seems BTXA does work better on the inflammatory state of the disease. We are now routinely injecting the sweat gland in these areas, and also focusing more on some occasional small papules which are the worst presentation of her HS now. The injection resolves the papules very well. The original sinus and fistula healed with monotherapy of botulinum toxin injection.

Botulinum toxin could be a cost-effective treatment for HS, even at Hurley stage III. Further large scale randomised controlled clinical trials need to be carried out to confirm the efficacy of botulinum toxin in the treatment of HS and to standardise treatment protocol.

## Patient's perspective

This is a life-changing treatment. We should let more patients with HS benefit from this botulinum toxin injection.

## Learning points

- Hidradenitis suppurativa (HS) could be well controlled by botulinum toxin injection.
- It is safe to inject botulinum toxin into the inflamed areas with HS.
- Even Hurley stage III HS could be amenable to botulinum toxin treatment.

**Correction notice** This article has been corrected to remove Figure 1. The image was provided by the patient and was consistent with the details of the wound in her records. It has been determined that the image was of a different patient. Its inclusion in the article was an honest error by all concerned.

**Contributors** WS: treating physician, collected the clinical data and wrote the manuscript. SS and AS: treating resident physicians, edited the manuscript. DRG: supervised the clinical management and critically revised the manuscript.

**Funding** The authors have not declared a specific grant for this research from any funding agency in the public, commercial or not-for-profit sectors.

**Competing interests** None declared.

**Provenance and peer review** Not commissioned; externally peer reviewed.

## REFERENCES

1 Napolitano M, Megna M, Timoshchuk EA, *et al*. Hidradenitis suppurativa: from pathogenesis to diagnosis and treatment. *Clin Cosmet Investig Dermatol* 2017;10:105–15.
2 O'Reilly DJ, Pleat JM, Richards AM. Treatment of hidradenitis suppurativa with botulinum toxin A. *Plast Reconstr Surg* 2005;116:1575–6.
3 Khoo AB, Burova EP. Hidradenitis suppurativa treated with Clostridium botulinum toxin A. *Clin Exp Dermatol* 2014;39:749–50.
4 Feito-Rodríguez M, Sendagorta-Cudós E, Herranz-Pinto P, *et al*. Prepubertal hidradenitis suppurativa successfully treated with botulinum toxin A. *Dermatol Surg* 2009;35:1300–2.
5 Heckmann M, Ceballos-Baumann AO, Plewig G, *et al*. Botulinum toxin A for axillary hyperhidrosis (excessive sweating). *N Engl J Med* 2001;344:488–93.
6 Yu CC, Cook MG. Hidradenitis suppurativa: a disease of follicular epithelium, rather than apocrine glands. *Br J Dermatol* 1990;122:763–9.
7 van der Zee HH, de Ruiter L, Boer J, *et al*. Alterations in leucocyte subsets and histomorphology in normal-appearing perilesional skin and early and chronic hidradenitis suppurativa lesions. *Br J Dermatol* 2012;166:98–106.