Prepubertal Hidradenitis Suppurativa Successfully Treated with Botulinum Toxin A

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Hidradenitis suppurativa (HS) is a recurrent scarring suppurative disease of the apocrine gland-bearing areas of the body that usually affects the intertriginous areas. Prepubertal HS is a rare condition; fewer than 2% of patients have onset of the disease before the age of 11.1 Management of HS, in children and adults, may be difficult because of its chronic nature and lack of response to standard therapeutic options, and there is little evidence of the efficacy of the different treatments in randomized clinical trials or after long-term follow-up.2–4 Among all therapies, botulinum toxin A (BTX) has been regarded as effective for isolated case reports of adult-onset HS.5 Recently, we succeeded in treating a case of prepubertal HS with BTX, which to our knowledge has not previously been described.

A 6-year-old girl with no relevant family or medical history was referred to the Department of Dermatology for evaluation of skin lesions that developed more than a year before and affected the patient’s daily activities because of pain. Physical examination revealed tender erythematous papules and solitary nodules involving only the groin, with remarkable symmetry (Figure 1). She was at the 75th percentile for weight and 70th for height. Her body mass index was 16.5 kg/m². She had no breast development and no pubic or axillary hair (Tanner stage 1 of sexual development), and no evidence of androgen excess was observed. An endocrinological examination yielded results within normal limits, and a bone age determination was also normal. A bacterial swab grew only coagulase-negative Staphylococci and Streptococcus species. Topical antibacterial medications (clindamycin and azelaic acid 15%) and oral erythromycin had been effective and resulted in remission, but early relapse occurred after stopping them. At aged 7, oral isotretinoin was initiated at a dose of 0.7 mg/kg per day but had to be discontinued after 4 months because of the development of painful deep-seated nodules and adverse effects. At 7 years, 5 months, a total dose of 40 mouse units of BTX (Botox; Allergan Pharmaceuticals Ireland, Westport, Ireland) was injected intradermally at 10 to 12 points over the elliptical area on each side. This procedure was performed with topical anesthesia and inhalation of nitrous oxide (50% nitrous oxide, 50% oxygen), with good tolerance. She experienced complete remission of her HS until 6 months later, when the first lesions reappeared. The relapse responded to a second treatment as well as they had to the initial treatment (Figure 2).

Early-onset HS may occur in the absence of endocrine abnormalities or androgen excess in prepubertal children.6 Nevertheless, the disease has also been reported in infants with congenital adrenal hyperplasia, obesity, or early puberty,1 so it is advisable to perform an endocrinological examination to rule them out. Many therapies are available for HS, but the benefits of these treatment options must be weighed against their side effects,
specially in childhood. Although medical treatments are usually enough to control early stages of the disease, once scarring appears, surgical interventions become the preferred mode of treatment. Intradermal BTX therapy may be a safe, well-tolerated, and effective alternative for young patients who develop mild to moderate HS showing no improvement after several medical therapies have been tried. BTX works at peripheral cholinergic synapses by temporarily blocking the evoked release of the neurotransmitter acetylcholine. These postganglionic sympathetic (cholinergic) nerve fibers stimulate eccrine sweat glands, so excessive sweating of focal hyperhidrosis can be reduced, and they might also stimulate apocrine sweat glands that have been implicated in the pathophysiology of HS. However, HS is now considered a primary disorder of the hair follicle, with subsequent inflammation and suppuration of the apocrine glands as a secondary phenomenon resulting from follicular occlusion. It has been suggested that BTX acts by reducing the production of apocrine sweat and limiting the tendency of follicular rupture, thus not allowing local inflammation and abscess formation. Nevertheless, recent studies have demonstrated the importance of adrenergic receptors localized in the apocrine sweat glands, indicating that other pathogenetic pathways may also be involved. It is likely that patients will require more than one treatment over time, because lesions tend to recur after an average of 6 to 10 months. Therefore, the necessity for optimizing BTX treatment is evident; further studies would be helpful in identifying the best candidates for BTX therapy.

References

Figure 1. Moderate hidradenitis suppurativa involving the groin (before treatment with botulinum toxin A).

Figure 2. Complete remission of the disease 6 months after treatment with botulinum toxin A.


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