CASE REPORT

Two patients with localised hyperhidrosis of the hand based on functional and structural abnormalities of sweat glands

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SUMMARY

A 14-year-old girl and a 30-year-old woman presented with localised hyperhidrosis on the dorsal hand and wrist, respectively, provoked by different stimuli such as physical activity and minor trauma to the skin. The skin was seemingly normal in both patients where an iodine–starch test revealed a well-demarcated area of hyperhidrosis. Following histopathological examination, the diagnosis was unilateral localised hyperhidrosis in both cases; one with normal histology and one with a nevus sudoriferous. Both patients were successfully treated with botulinum toxin type A. The 30-year-old woman additionally used low-dose propantheline bromide periodically and experienced long-term remission on this therapy. Hyperhidrosis may embarrass and interfere with patients' school and careers, and it is therefore important to tailor an effective individual treatment.

BACKGROUND

Nevoid focal hyperhidrosis, also known as a nevus sudoriferous or eccrine nevus, is a rare benign skin hamartoma composed of an increased number or size of eccrine glands. Less than 50 cases have been reported in the world literature. In other cases, focal hyperhidrosis is caused by a functional hamartoma with sweat glands in the affected area being excessively sensitive.¹ In daily clinic, the extent of a hyperhidrotic skin area can be visualised by the iodine–starch test (Minor's test) used to evaluate sudomotor function: an iodine solution is applied to the affected skin area. Once dry, the skin is dusted with starch, and sweat will make starch and iodine combine turning into a dark-violet colour, thereby allowing visualisation of the hyperhidrotic skin area.

CASE PRESENTATION

Patient 1: A 14-year-old otherwise healthy girl was referred to our department due to localised hyperhidrosis dorsolaterally on the left hand, which had been present since the age of 6. Hyperhidrosis was worse during wintertime, especially when wearing gloves, and could also occur at night. The patient experienced episodes of hyperhidrosis at least twice a week, each time with a duration of several hours. She complained of trouble writing on paper, as droplets of sweat would slide down and soak it. Bandaging the area only further provoked

hyperhidrosis. She managed symptoms by wiping off the sweat with a cotton cloth. On examination, a discrete, pale red area measuring about 3×5 cm was seen on the dorsum of her left hand.

Patient 2: A 30-year-old otherwise healthy woman was referred to our department due to localised hyperhidrosis on the right wrist and adjacent dorsal part of her hand, which had been present since the age of 7. Episodes of hyperhidrosis would occur two to three times weekly with duration of 20 min to 1 hour. Hyperhidrosis was provoked by stress, heat, hot water, physical activity and after minor trauma to the skin such as an irritating sleeve. She worked as a dental assistant and had some practical problems at work, when sweat was dripping from this specific area. She had previously tried to use a bandage, but this was not a viable option for hygienic reasons. Seemingly, the affected skin area was not different from the surrounding skin, although she had noticed that the area would become slightly erythematous during episodes of hyperhidrosis.

INVESTIGATIONS

Patient 1: Sweat droplets were emerging from the affected skin and could be seen running down her hand (see [figure 1A](#)). Iodine–starch test was performed and revealed an approximately 5×5 cm demarcated area of hyperhidrosis on the dorsum of the hand (see [figure 1B](#)). A punch biopsy showed an increased amount of eccrine sweat glands with normal appearance, equivalent to a nevus sudoriferous (see [figure 2A](#)).

Patient 2: Iodine–starch test was performed and revealed a palm-sized demarcated area of hyperhidrosis on the patient's right wrist and dorsal hand (see [figure 3](#)). A punch biopsy showed an estimated normal amount of eccrine sweat glands (see [figure 2B](#)).

TREATMENT

Patient 1 initially tried topical aluminium chloride and iontophoresis with no effect. She was then successfully treated with intralesional injections of botulinum toxin (100 IU Botox) and is currently awaiting her second treatment.

Patient 2 initially tried topical aluminium chloride with no effect. She was then successfully treated with systemic propantheline bromide (15 mg two times a day) and yearly intralesional injections of botulinum toxin (varying between 25–100 IU Botox).

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Figure 1 Nevus sudoriferous (of patient 1). Sweat can be seen on the dorsum of the patient's left hand (A), and the hyperhidrotic area is further visualised by iodine–starch test (B).

OUTCOME AND FOLLOW-UP

Based on the clinical symptoms and histopathology, a diagnosis of nevus sudoriferous and unilateral localised hyperhidrosis was made in patient 1 and patient 2, respectively.

DISCUSSION

Hyperhidrosis is divided into two main categories: primary and secondary hyperhidrosis. Primary hyperhidrosis is idiopathic and mostly localised. It usually occurs bilaterally and affects the head, axillae, trunk, palms, soles and anogenital area. Secondary hyperhidrosis is often compensatory and asymmetrical/regional due to denervation or generalised due to medications or an underlying medical condition.

In the case of a nevus sudoriferous, however, hyperhidrosis can occur unilaterally and anywhere on the body. Seemingly, the nevus sudoriferous is most often located on the forearm (50% of cases) but may also be present on the back and trunk, as well as on the forehead and extremities.^{2–10} Clinical manifestations range from no epidermal changes^{11–13} to dyspigmentation, linearly arranged papules, depressed brown patches or a centrally depressed nodule surrounded by a slightly scaly border.^{3–6 14–16}

Lesions typically appear during childhood or adolescence, however, congenital lesions as well as symptoms starting at the age of 79 have been reported.^{3 14 17 18}

Although the nevus itself is harmless, it typically has a negative impact on patients' quality of life, as they may find it embarrassing, and it can be difficult to wear clothes. It may also interfere with patients' school or career, as was the case in our patients.

Figure 2 (A) Skin biopsy (of patient 1) with an increased amount of eccrine sweat glands with normal appearance, equivalent to a nevus sudoriferous. (B) Skin biopsy (of patient 2) with an estimated normal amount of eccrine sweat glands. (H&E stain, original magnification $\times 25$).

Figure 3 Unilateral localised hyperhidrosis (of patient 2) visualised by performing an iodine–starch test.

Treatment usually consists of application of topical aluminium chloride, iontophoresis, intralesional injections of botulinum toxin or surgical excision of the nevus.^{19 20} Intralesional administration of botulinum toxin, which prevents the release of acetylcholine at the neuromuscular junction, is very effective in treating localised hyperhidrosis although requiring serial reinjections. Successful treatment with topical glycopyrrolate has also been reported.²¹ Due to the anatomic location in these two cases, endoscopic thoracic sympathectomy could be an additional option. However, we think this should be the last therapeutic option due to potential side effects including a more generalised compensatory hyperhidrosis. Also, there are examples of patients having this procedure performed without any effect on the frequency of sweat attacks. Possibly, sudomotor nerves may have only a minor role in nevus sudoriferous.

In general, it may not seem justified to treat focal hyperhidrosis with systemic anticholinergic drugs such as propantheline bromide (which was used by patient 2 in our case report), due to the possibility of local treatment as well as the risk of side effects of systemic acetylcholine receptor blockage. However, our

Patient's perspective

- ▶ Patient 1: "My hand bothers me a lot. It is constantly sweating during winter, so that for example in gym class, the floor or my clothes get wet. In school, the table, books and so on get wet during classes. In the night-time, when the hand is sweating, the sheets get soaked. In general, it is very discomforting that my hand sweats so much. Now I have tried a Botox treatment and it has helped me 100%, the hand no longer sweats."
- ▶ Patient 2: "Each time I started sweating, I would be embarrassed and sad about it. At work, I had to be very careful: Whenever I could feel heat on my wrist, I would rush out and put on an elastic hand bandage."

Learning points

Localised hyperhidrosis:

- ▶ Is a rare condition.
- ▶ Can be caused by a nevus sudoriferous or a functional hamartoma.
- ▶ Can be successfully treated with botulinum toxin.
- ▶ Can greatly impair daily functioning and quality of life.

patient was very satisfied with low-dose propantheline bromide (15 mg two times a day), as she experienced very few side effects and preferred an oral drug rather than injections with botulinum toxin, which she found painful.

In summary, localised hyperhidrosis can be treated in a variety of ways. We recommend aluminium chloride and botulinum toxin as the treatment of choice. All available options should be carefully tailored to the individual patient, as the efficacy and side effect profile is individual.

Contributors BHK was the main contributor to all aspects of planning and writing of the article, including clinical images. KHL contributed to the histopathological images and the description of these. KAUP contributed to writing the part regarding patient 2. AB contributed to planning and writing/editing all parts of the article. All of the authors certify that they will take public responsibility for the contents, have contributed substantially to the drafting and have approved the final version.

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