

Efficacy of botulinum toxin for a congenital eccrine naevus

Editor

Congenital eccrine naevus is a rare clinical entity. Fewer than 20 cases had been reported, and only three cases were congenital.^{1,2} We present a child with congenital localized hyperhidrosis on the forearm. To the best of our knowledge, this is the first congenital eccrine naevus successfully treated with botulinum toxin type A.

A 12-year-old girl with no significant past medical history presented with a history of localized excessive sweating on the right forearm from birth. The episodes were twice a week and were initially confused by their parents with enuresis. At 6 years of age, the episodes began more frequently, compromising her school activities. The episodes occurred daily, were preceded by a paresthetic sensation on the area and were precipitated by heat, physical activity and specially by the muscular strain of handwriting. Physical and neurological examinations were normal. Epidermic changes were not observed in the hyperhidrotic area.

A provocation test with exposition to heat for 30 min confirmed a hyperhidrotic area of 5 × 4 cm localized on the ulnar eminence of the right wrist. Sweat dripped from the area (fig. 1). An incisional biopsy specimen showed a proliferation of eccrine glands of the deep dermis and hypodermis (fig. 2).

Topical treatment with aluminium chloride 20% was unsuccessful. Surgical extirpation was discarded due to the size of the lesion. Considering the interference with her school activities and according to her mother's previous consent, treatment with botulinum toxin type A (Botox Allergan, USA) was performed. Botulinum toxin type A was diluted in 4 mL of saline solution 0.9%, with 5 U per intradermic injection, at intervals of 0.5 to 1 cm.



fig. 1 Localized hyperhidrosis on the right wrist.

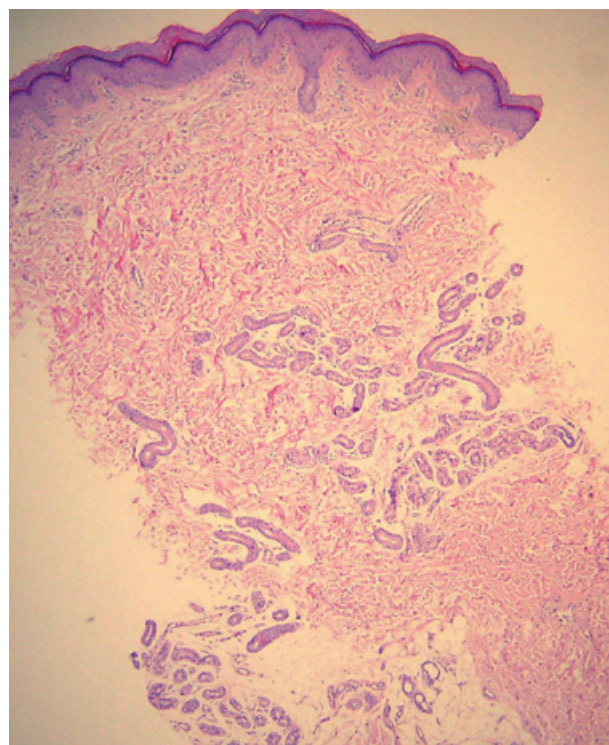


fig. 2 Histological examination showed an increased number of eccrine sudoriparous glandular ducts and acines (haematoxylin and eosin).

Up to date, 1 year after, a significant decrease of sweat episodes to once a month, and an improvement in our patient's quality of life was achieved.

This case is the first congenital eccrine naevus successfully treated with botulinum toxin, according to our literature revision. Eccrine naevus (EN) is a hamarthome characterized histologically by increased number and/or size of the eccrine glands, without vascular proliferation. It should be differentiated from eccrine angiomatose hamarthome and from sudoriparous angiome. Eccrine angiomatose hamarthome is a frequent pathology that presents a vascular proliferation associated with hyperplasia of the sudoriparous glands.³ Hypertrophy of the eccrine glandular component with vascular proliferation is named sudoriparous angiome. In this case, the absence of vascular proliferation, confirmed the diagnosis of a pure eccrine naevus, with fewer than 20 cases reported.^{2,4}

As seen in this patient, EN often presents as a localized hyperhidrosis without overlying skin abnormalities and respond to physiological stimuli of perspiration.¹ Half of cases are localized on the forearm. Kopera and Soyer⁵ suggested that this preferential localization on the forearm would be phylogenetically related to the lemur catta antibrachial organ, formed by sudoriparous glands to delimit its territory.

Treatment may be difficult, specially when hyperhidrosis has a significant impact on the patient quality of life. In this case, hyperhidrosis was unresponsive to topical therapy, and according to cholinergic innervation of eccrine glands, treatment with botulinum toxin type A was successfully performed. Similar treatment of an acquired eccrine naevus was recently reported.⁶

Our case is unusual in that to the best of our knowledge, is the first congenital eccrine naevus treated with Botox and highlights the positive outcome of this option when surgical procedure is not considered.

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Erythema gyratum repens-like eruption in mycosis fungoides: is dermatophyte superinfection underdiagnosed in cutaneous T-cell lymphomas?

Editor

Mycosis fungoides (MF) is the most frequent T-cell lymphoma. It can mimic either malignant or benign dermatoses. We report a particular form of mycosis fungoides mimicking erythema gyratum repens (EGR).

A 77-year-old man had been followed at our department for 2 years because of stage III MF (T4N1M0B0).¹ The patient had received various therapies, including chlormethine, carmustine and methotrexate, which all had failed. Recently, a polychemotherapy was initiated with vincristine, cyclophosphamide and prednisone. The cutaneous lymphoma was partially controlled by the treatment with a few residual patches, but the patient presented at follow-up visit with a 3-week duration eruption involving the abdomen, the back and the buttocks (fig. 1). The lesions were pruriginous, slightly desquamative with a typical double-ring. This eruption was clinically consistent with a diagnosis of EGR. A cutaneous skin biopsy and a mycological sample were obtained because of our doubt on the association of typical MF with the rare paraneoplastic syndrome: EGR. Histopathology showed a characteristic pattern of mycosis fungoides associated with the presence of mycelia on periodic acid–Schiff staining (fig. 2). *Trichophyton rubrum* was isolated in cultures. The diagnosis of EGR was ruled out based on these results, and the patient was considered to have a MF aggravated by an extensive dermatophytosis. A treatment was begun with systemic terbinafine 250 mg daily associated with a daily cutaneous application of ketoconazole 2% in cream for 15 days. The annular lesions improved rapidly, but the cutaneous lymphoma worsened with the apparition of a significant number of Sezary's cells on blood examination and the extension of typical cutaneous MF lesions (stage IV). The patient was proposed to receive photochemotherapy treatment.

The classical lesions of MF are patches or plaques. The annular aspect of MF is frequently seen in the plaques, often with central clearing. On one hand, some atypical variants of MF are nowadays well recognized such as folliculotropic (with or without mucinosis), pagetoid reticulosis and granulomatous slack skin types² as well as hypopigmented or hyperpigmented variants in black skin patients.^{3,4} On the other hand, in many previously published cases, MF can simulate other dermatoses.⁵ Particularly, several cases of MF mimicking annular erythema have been reported in the literature. In these cases, mycology was negative and histology was typical of MF.^{6,7} Another form of annular MF has been described in children as 'lymphomatoid annular erythema'.⁸ In our case, the patient presented with an annular eruption consistent with EGR. The association of MF with true EGR has never been reported to the best of our knowledge. The skin biopsy sample of the edge of a ring showed a typical MF infiltrate associated with the presence of a fungus (fig. 2), suggesting a diagnosis of MF lesions associated with *T. rubrum* infection. This EGR-like eruption may have been caused by the combined features of fungal infection and epidermotropic malignant T-cell infiltrate.