

Idiopathic Unilateral Localized Hyperhidrosis

Sir,

Unilateral localized hyperhidrosis is an uncommon condition with diverse causes. Lesions affecting any part of the sympathetic nervous system are often encountered as the cause of the disease and in a few cases enlarged sweat glands have been shown; these cases have been regarded as a type of eccrine naevi. There are also some cases in which the underlying pathology cannot be determined, and these are considered idiopathic. We describe here another case of idiopathic unilateral localized hyperhidrosis; a disorder with an aetiology still waiting to be clarified.

CASE REPORT

A 45-year-old male presented with a 4-year history of increased sweating limited to the right side of his face and scalp (Fig. 1). Severe attacks of sweating lasting for about an hour were precipitated by increased environmental temperature, emotional stress, eating, drinking, exercise and sometimes without an obvious stimulus. There was no preceding history of trauma, parotid disease, neurological, ophthalmological or otolaryngeal abnormalities. Hyperhidrosis was not accompanied with flushing, excessive salivation, lacrimation, vasodilatation, orthostatic hypotension, causalgia or headache. He was in good health and a physical examination was unremarkable except for profuse sweating on the right side of his face and moistness of the hair over the right side of the scalp. Large drops of sweat dripped continuously from the forehead and nose and the area of hyperhidrosis was sharply demarcated with an abrupt cessation of sweating at the midline. The starch-iodine test performed on corresponding areas was immediately and strongly positive on the right, with a much slower reaction on the left (Fig. 2). The other half of his face and scalp, and also his trunk and extremities sweated within normal limits, as shown by a physical examination performed after strenuous physical exercise, which revealed visible sweat droplets on both sides of the face and on other parts of the body.

Neurological and otolaryngeal examinations revealed no abnormal findings. There was no accompanying sensory or motor neuropathy and no associated autonomic dysfunction was observed. His blood pressure and pulse rate were within the normal range. Thermoregulation was normal and the patient had no gastrointestinal symptoms or abnormal patterns of defecation or micturition. The typical features of Horner's syndrome other than ipsilateral anhidrosis; such as miosis, ptosis and enophthalmos, were not present. Complete blood

count and hepatic and renal function tests were found to be in the normal range. Blood glucose was in the normal range and tests for syphilis were negative. X-rays of the chest and magnetic resonance imaging of the brain and brainstem were normal. Skin biopsies taken from the affected and symmetrically opposite normal area of the skin were examined for any eccrine gland abnormalities and were found normal. He was diagnosed as having idiopathic unilateral localized hyperhidrosis and amitriptyline in a dosage of 10 mg/day combined with topical aluminium chloride cream (20%) was started as treatment. One month later amitriptyline dosage was increased to 20 mg/day but no significant improvement in the hyperhidrosis was achieved.

DISCUSSION

Idiopathic unilateral localized hyperhidrosis occurs mainly on the face and upper extremities of otherwise healthy individuals (1–5). Profuse sweating, usually in a sharply demarcated area, is most commonly precipitated by heat and in some cases also triggered by emotional stress and gustatory stimulation. Unilateral localized hyperhidrosis has rarely been reported in the literature during the last 50 years and the pathogenesis of the disease still remains to be clarified.

Unilateral localized hyperhidrosis has been shown to occur with lesions involving any part of the sympathetic nerve pathway, such as syringomyelia, intramedullary glioma, vertebral osteoma, head and spinal cord injury, structural lesions of the hypothalamus and spinal cord infarction (6–8). In this group of patients, sweating is usually seen without a stimulus and may be transient. Unilateral hyperhidrosis has been reported to occur contralateral to acute cerebral infarction and it has been attributed to the disruption of a pathway of cortical origin, inhibitory to contralateral sweating. Some authors suggest that paroxysmal unilateral hyperhidrosis may be regarded as a sign of poor prognosis in stroke due to its occurrence in patients with relatively large strokes affecting superficial and deep subcortical structures (8). Unilateral hyperhidrosis has been reported secondary to bronchial carcinoma suggestive of neoplastic involvement of the ipsilateral sympathetic chain. In these rare cases paroxysmal profuse sweating is noted even during sleep (6,



Fig. 1. Profuse sweating on the right side of the face with an abrupt cessation at the midline

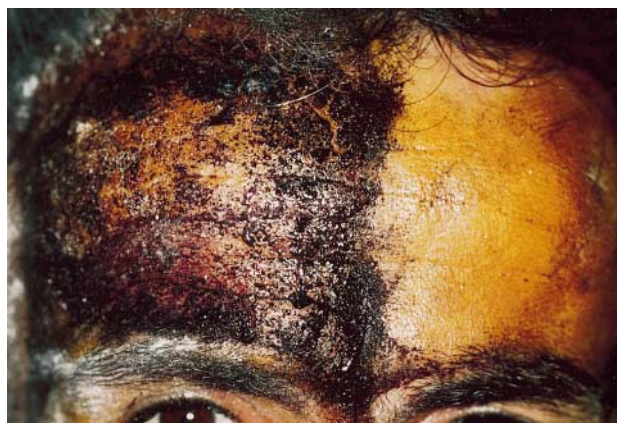


Fig. 2. The starch-iodine test, showing an immediate strong reaction on the right side of the face.

7). In our patient there was no history of any injury to the head and neck or parotid disease, physical examination revealed no neurological signs, no associated Horner's syndrome or other autonomic dysfunction was observed and magnetic resonance imaging of the brain and brainstem were normal.

There are few reports demonstrating the presence of enlarged sweat glands in the affected skin of patients with localized hyperhidrosis and the lesions have been considered as variants of the pure anatomical eccrine naevi or as functional naevi which showed secondary hypertrophy of the glandular elements (1, 9, 10). But this finding has not been reported in most of the patients with localized hyperhidrosis, and skin biopsies in our patient failed to reveal such findings.

According to some authors (4), localized hyperhidrosis may be caused by a non-demonstrable disturbance of autonomic nervous system, but the reason for this rare condition remains to be elucidated.

Because of the limited number of cases reported, there is no standard therapy for idiopathic unilateral localized hyperhidrosis and the condition often proves resistant to therapy. Sweating may partially be controlled by treatments with anticholinergic drugs, sedatives, tranquilizers, calcium-channel blockers, astringents, topical antiperspirants, iontophoresis and surgical sympathectomy. However, these treatments have various side-effects and topical therapies tend to cause contact dermatitis (1, 5, 10). In our patient treatment with amitriptyline in a dosage of 20 mg daily combined with topical aluminium chloride cream 20% was not found to be effective.

REFERENCES

1. Cunliffe WJ, Johnson CE, Williamson DM. Localized unilateral hyperhidrosis – a clinical and laboratory study. *Br J Dermatol* 1972; 86: 374–378.
2. Verbov J. Unilateral hyperhidrosis over face and scalp. *Br J Dermatol* 1974; 90: 470.
3. Van De Kerkhof PCM, Den Arend JACJ, Bousema MT, Stolz E. Localized unilateral hyperhidrosis. *Br J Dermatol* 1987; 117: 779–782.
4. Köse O, Baloglu H. Idiopathic unilateral circumscribed hyperhidrosis. *Int J Dermatol* 1997; 36: 198–212.
5. Takase Y, Tsushimi K, Yamamoto K, Fukusako T, Morimatsu M. Unilateral localized hyperhidrosis responding to treatment with clonazepam. *Br J Dermatol* 1992; 126: 416.
6. Pleet DL, Mandel S, Neilan B. Paroxysmal unilateral hyperhidrosis and malignant mesothelioma. *Arch Neurol* 1983; 40: 256.
7. Middleton WG. Bronchial carcinoma with pleural spread causing unilateral thoracic hyperhidrosis. *BMJ* 1976; 2: 563.
8. Labar DR, Mohr JP, Nichols FT, Tatemichi TK. Unilateral hyperhidrosis after cerebral infarction. *Neurology* 1988; 38: 1679–1682.
9. Bedi TR, Bhutani LK. Unilateral facial hyperhidrosis. *Dermatologica* 1974; 149: 374–378.
10. Ruiz de Erenchun F, Vazquez Doval FJ, Contreras Mejuto F, Quintanilla E. Localized unilateral hyperhidrosis: eccrine nevus. *J Am Acad Dermatol* 1992; 27: 115–116.

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Ayşe Boyvat, Gamze Pişkin and Hatice Erdi
Department of Dermatology, Ankara University School of Medicine, İbni Sina Hospital, Samanpazari-Ankara, TR-06100, Turkey.

Pityriasis Rubra Pilaris: A Retrospective Analysis of 43 Patients

Sir,

Pityriasis rubra pilaris (PRP) is a rare skin disease of unknown aetiology. Since 1980 we have seen 43 patients with this diagnosis. This study describes these patients and the course of their disease, based on a retrospective analysis.

MATERIALS AND METHODS

Patients

The study comprised 43 patients (12 females and 31 males). Their age range at the start of disease was 1–77 years (Fig. 1). Forty-one of the patients were admitted for investigations and treatment, while only 2 had outpatient treatment. Nine women and 24 men (mean age 58 years) had adult onset of disease (>15 years of age) and 3 girls and 7 boys (mean age 8 years) had juvenile onset (<15 years of age).

Diagnosis

The clinical picture is somewhat different for the various types of PRP. In its classical form, the skin eruption most often starts in the face and associated areas and then spreads within a few weeks to the rest of the body, leaving only small islands of unaffected skin (Fig. 2). The eruption is erythematous and scaling with follicular plugging. The palms and soles are yellowish and hyperkeratotic. Within 2–3 months erythrodermia may develop. Some patients also develop ectropion (1).

In our material it was not possible to distinguish between the

various types of PRP described (2), because the clinical information in the medical reports was not sufficiently accurate.

Most patients were diagnosed from the clinical picture and the diagnosis was verified histologically. In 8 patients the diagnosis could not be verified by our pathologist. However, the clinical pictures were so characteristic that the diagnosis was maintained. Five of those patients were females. This means that in 42% of the females, the diagnosis of PRP could not be verified histologically, whereas this was the case in only 10% of the males ($p < 0.05$).

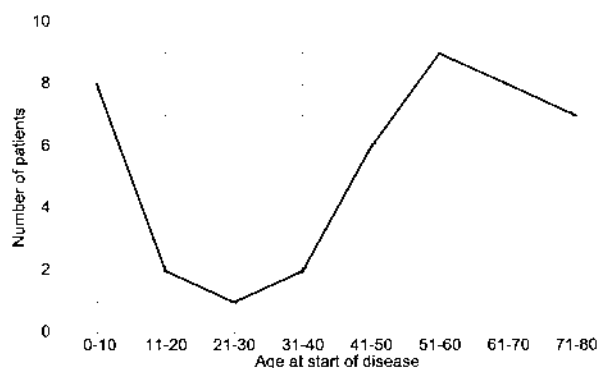


Fig. 1. Age distribution at the start of the disease.