

Correspondence

Allergic contact dermatitis due to diclofenac applied with iontophoresis

A 37-year-old woman presented with itchy, erythematous, oedematous and vesicular lesions on the lumbar region. Ten days earlier she had started a twice weekly treatment of iontophoresis with Voltaren® ampoules (diclofenac) to treat lumbar pain. She had not previously used diclofenac either systemically or topically. The eczematous reaction disappeared promptly after discontinuation of iontophoresis treatment and application of mometasone furoate cream for 7 days. Patch tests with SIDAPA (Italian Society of Allergological, Professional and Environmental Dermatology) standard series and with diclofenac 1% and 2% (aq.) were performed using AI-tests (Imeco ab, Södertälje, Sweden). Readings were made after 2 and 4 days (D2 and D4, respectively). Patch testing gave an intensely positive reaction (++++) to diclofenac at D2 and D4 at both concentrations. Positive results (++) were also recorded to colophony and *Myroxylon pereirae* resin at D2 and D4. Photopatch tests with diclofenac and patch tests with excipients contained in Voltaren® ampoules were negative.

Diclofenac is a nonsteroidal anti-inflammatory drug (NSAID) of the acetic acid group, which is available as oral, topical and parenteral preparations. Side-effects associated with topical application have been reported rarely, despite the widespread use especially in Mediterranean countries, and consist of allergic contact dermatitis caused by the application of eyedrops and gel preparations.^{1–6} Positive patch test reactions have been also elicited in some patients who developed a maculopapular rash after systemic administration of the drug.⁷ Diclofenac does not appear to cross-react with buprenorphine, another NSAID which frequently causes contact allergy.⁸ Buprenorphine and diclofenac both have a benzene ring in their structure but have a different chemical group on the alkyl chain, which may explain the apparent lack of cross-sensitization.

Iontophoresis is a well-known technique aimed to the transdermal delivery of ionic therapeutic agents by the application of a low-energy electric current.⁹ In our patient, the release of high concentrations of diclofenac into the skin through iontophoresis might have augmented the risk of sensitization and might have induced a rapid development of the eczematous reaction.

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Accepted for publication 3 June 2003

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An elderly patient with mycosis fungoides successfully treated with chronic low-dose oral etoposide therapy

The management of advanced stage mycosis fungoides (MF) is difficult, especially in elderly cases. Here we report an elderly patient with clinical stage III MF who was successfully treated with oral administration of etoposide for 5 years.

The patient was a 75-year-old Japanese male who was admitted to our hospital with a 2-year history of erythrodermic skin lesions in August 1992. Physical examination revealed dark red-pigmented plaques, erosions, and poikiloderma, which covered his entire body surface. Histological examination of the lesional skin revealed atypical CD4+ T cells infiltrating the upper dermis with epidermotropism, however, no Sezary cells were detected in peripheral blood. Bilateral axillary and inguinal lymphadenopathy were noted, which showed no specific histological signs upon lymph node biopsy. No other visceral involvement was detected. Thus, a diagnosis of erythrodermic MF, pT4N1M0, clinical stage III was made.

He was first treated with the combination of retinoid-PUVA and electron beam (EB) therapy for 10 weeks, but showed no clinical improvement. We then chose a combination of intravenous etoposide 150 mg/day for the 1st day and oral prednisolone 30 mg/day for the 1st, 2nd and 3rd day of every week for 9 weeks. After this therapy, his skin became almost free of eruptions, and lymph nodes became impalpable. We observed no significant adverse effect with this therapy. He was discharged, and then we started chronic low-dose oral etoposide therapy, oral etoposide 25 mg/day for 21 days every 4 weeks. After the first course of this therapy, his skin lesions completely cleared up. We continued this therapy for 60 months. No apparent clinical recurrence was noted for 36 months until increasing numbers of plaques and erosions recurred. In April 1998, he was admitted to our hospital again, and he was treated with the combination of intravenous etoposide 200 mg/day for the 1st day and oral prednisolone 30 mg/day for the 1st, 2nd and 3rd day of every week for 3 weeks. Although a slight response was observed, the patient was affected by MRSA enteritis in May 1998. We immediately stopped etoposide and started to give oral vancomycin, but he developed septicemia and died of disseminated intravascular coagulation (DIC) in July 1998. Autopsy revealed that tumour cell infiltration was restricted to the skin, and no visceral involvement was detected.

There exist difficulties associated with treatment in the advanced stages of MF, especially in elderly cases. Although many combination or single agent chemotherapies have been reported as being effective in the treatment of MF, no consensus has been achieved for the preferred therapy.¹ Combination chemotherapies that have a higher rate of responsiveness tend to cause significant adverse effects, such as myelosuppression, cardiotoxicity, which are often problematic or even fatal in the elderly patients.

Chronic oral administration of etoposide has been used in the therapy of refractory neoplasia including lymphomas during the 1990s.² The administration of this drug is possible on outpatient basis, and it is surprisingly effective in the patients heavily pretreated with etoposide-containing combination chemotherapies.³

Moreover, oral etoposide has been reported to be a useful therapy in the treatment of elderly patients with lym-

phoma.⁴ This is the first case report of an elderly MF case successfully treated with oral etoposide over a period as long as 5 years. We suggest that oral etoposide is a useful palliative chemotherapy especially for elderly MF patients, with an extended period of well tolerated efficacy, minimal adverse effects and better quality of life for the patients.

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Accepted for publication 13 June 2003

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Cutaneous hyperpigmentation during therapy with hydroxychloroquine

We report a 48-year-old female systems analyst who presented to the rheumatology clinic with bilateral metacarpophalangeal joint arthritis, facial erythema, Raynaud's phenomenon, carpal tunnel syndrome, and a history of two second trimester miscarriages. Her antinuclear antibodies (tested on rat liver) were detectable to a dilution of 1 in 2560, with a positive anti-U1 RNP antibody, supporting a diagnosis of mixed connective tissue disease. Anticardiolipin antibodies were negative, and DNA binding, C3 and C4 complement levels were normal.

The patient was advised about general photo-protection, and specific treatment was initiated with hydroxychloroquine (HCQ) 200 mg twice a day, producing a good therapeutic response within 2 months. After 1 year, the patient presented to the joint dermatology and rheumatology clinic (in August 2002) with a 3-month history of striking slate grey macular pigmentation affecting her face, neck, trunk, axillae and posterior thighs (Fig. 1a and b), thereby involving both sun-exposed and sun-protected sites. The pigmentation did not involve palmar creases or the buccal mucosa, and ophthalmic examination and testing was unremarkable. In addition, there was no history of any preceding inflammation, ingestion of tetracyclines, amiodarone, psychiatric drugs, application of

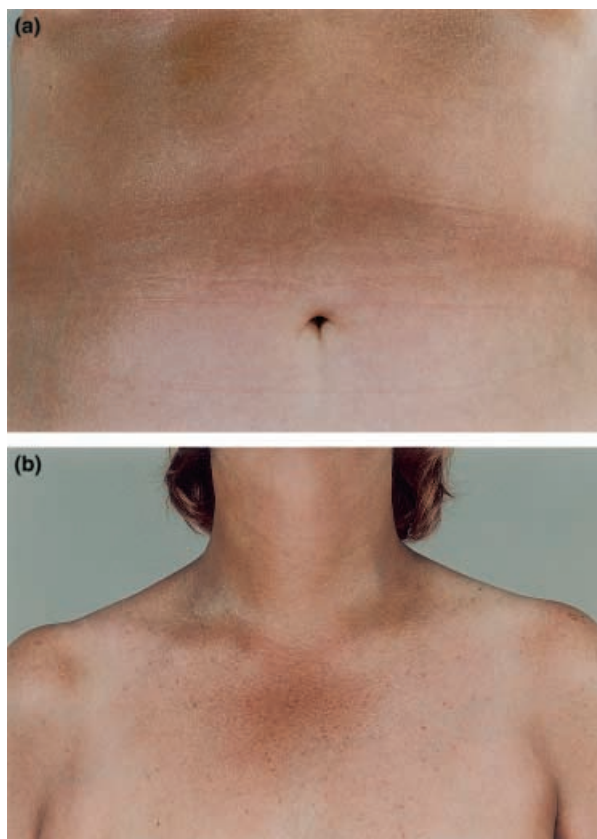


Figure 1 Striking slate grey macular pigmentation across the upper abdomen (a) and chest/neck (b).

hydroquinone, and on examination no clinical features to suggest arsenic ingestion.

Serum levels of ferritin, lead and silver were normal, as were the serum kidney, liver and thyroid function tests, glucose and full blood count. Histology of an incisional skin biopsy demonstrated marked pigmentary incontinence, a mild chronic inflammatory infiltrate and some basal cell degeneration. The patient has been unable to reduce the HCQ below 400 mg daily because of her joint symptoms, although her hyperpigmentation has improved recently in the winter months.

The close temporal relationship in this case between treatment with HCQ and appearance of the hyperpigmentation suggests that this antimalarial drug is the most likely cause. Although mucocutaneous hyperpigmentation is said to affect 10–25% of patients taking the major antimalarials (HCQ, chloroquine, mepacrine and mefloquine) for more than 3 months,^{1,2} our Medline search identified only two published reports of hyperpigmentation attributed to HCQ: one of cutaneous³ and one of gum⁴ pigmentation. Given the lack of published evidence for this apparently common side effect, we report the present case to illustrate this adverse reaction to HCQ.

Hyperpigmentation caused by other antimalarials was first reported during World War II among US soldiers in the Pacific who were treated with quinacrine.⁵ Since then many reports have documented pigmentary problems following treatment with all antimalarials, particularly amongst Caucasians, and most frequently causing a bluish (or occasionally grey to black) pigmentation. The hard palate is the commonest site, with pigmentation of the face, limbs (including pretibial), trunk and nails also reported.² The biochemical basis for the pigment is unknown, although it is usually reported to be reversible, fading within 2–6 months of discontinuing the drug.¹ Antimalarial drug-induced hyperpigmentation has also been suggested as a marker for patients at risk of ocular side effects.

In summary, our case illustrates cutaneous hyperpigmentation that developed during therapy with HCQ. Although a common adverse effect of antimalarial drugs in general, the lack of other similar cases reported with HCQ suggests that it may be less common with HCQ than with other antimalarial agents.

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Accepted for publication 23 June 2003

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Acquired primary cold contact urticaria after Hymenoptera sting

Cold urticaria is a heterogeneous group of manifestations defined as an urticarial reaction or angioedema on exposure to cold. In 96% of the patients the disease is idiopathic. The secondary form has been associated with infections (Epstein–Barr virus, *Mycoplasma pneumonia*, syphilis), neoplastic or immunologic diseases (cryoglobulinemia, Waldenstrom's disease, C2 and C4 defects).¹ Acquired primary cold contact urticaria has been rarely reported

after insect¹ and hymenoptera² stings, after jellyfish stings,³ and during hymenoptera venom desensitisation; localized cold urticaria has been reported at the site of immunotherapy injections of pollen extracts.⁴ Prevalence of cold urticaria has been estimated to be 1–3%. Although there are no published data regarding prevalence in Greece, daily clinical practice suggests a similar frequency.⁵ Prevalence of hymenoptera venom sensitization without clinical manifestations in Greece, has been reported to be 29%.⁶

We report four cases of acquired primary cold contact urticaria, associated with a prior hymenoptera insect sting.

Four young women presented with a first report of cold urticaria at the Allergy Department of A. Sygros Hospital, between 1997 and 2001 (Table 1). They had all developed pruritic fleeting wheals on the trunk and extremities and generalized erythema accompanied by a burning skin sensation, 15 min after entering sea water for swimming. Patients 2 and 4 also reported angioedema of the lips and eyelids. Symptoms remitted 10–40 min after leaving the water, except for the angioedema which lasted for about 12 h. All patients reported a 24–48-h-old hymenoptera (three honey bee and one common wasp) sting, which presented with only moderate local reaction. None of the patients received therapy for the stings. There was no report of viral or bacterial infection, systemic disease or history of any kind of drug intake. All patients reported hymenoptera stings in the past without systemic symptoms.

All patients were evaluated by ice cube provocation test (3, 6 and 10 min) and cold arm bath provocation test (15 min in 8 °C water). Laboratory evaluation included complete blood count, erythrocyte sedimentation rate, c-reactive protein, anti-nuclear antibody, rheumatoid factor, antistreptolysin-O (ASTO), immunoglobulin IgG, IgM and IgE levels, cold agglutinins, cryoglobulins, cryofibrinogen, total complement and C2, C3, C4, C1-

esterase inhibitor levels, syphilis, hepatitis B and C, borreliosis, mononucleosis and HIV serologies, anti-a-1-antitrypsin, thyroid hormones and thyroid stimulating hormone. Skin prick tests were performed with common food and inhalant allergens and specific IgE to hymenoptera venom (honey-bee, common and paper wasp, *Dolichovespula maculata* and *D. arenaria*) was measured (CAP System, Pharmacia, Uppsala, Sweden). Follow-up evaluation included ice cube and cold arm bath tests every 3 months and laboratory tests every 12 months for 2 years.

Patients displayed positive ice cube (at 3 min) and cold arm bath provocation tests during the first 6 months. From the ninth month on, the time needed for the tests to become positive was significantly extended. Positive ice cube test reactions were obtained after 6 min for patients 1 and 4, and 9 min for patient 2. The test was negative for patient 3. In all patients the provocation tests were negative 12 months after presentation. The patients were counselled to avoid swimming and exposure to cold. No antihistamines or other drug treatment was administered. Patients reported symptoms after cold exposure for several months (Table 1). Levels of total and specific IgE are also listed in Table 1. Laboratory tests were within normal limits except for patient 3 who presented a positive rheumatoid factor, marginally raised c-reactive protein and positive anti-thyroid antibodies as expected. Prick tests were negative. All patients remained without clinical manifestations for the next 12 months and there was no new hymenopteran sting during that time either.

Acquired primary cold contact urticaria has been suggested to be associated with insects stings including hymenoptera.^{1,2} We believe the four cases reported to have been triggered by hymenoptera sting. Although there is no solid evidence there are a number of indications that point out in this direction. These include: (a) the time interval between the hymenoptera sting and the onset of symptoms. In all cases the cold urticaria developed a few days after the

Table 1 Patient characteristics.

	Patient 1	Patient 2	Patient 3	Patient 4
Age (years)	18	37	29	22
Number of episodes	5	2	4	2
Medical history	Brucellosis when 13 years old	Unremarkable	Hypothyroidism due to Hashimoto's disease since age 18 years	Rheumatoid arthritis since age 22 years
Family history	Unremarkable	Unremarkable	Father with chronic urticaria	Unremarkable
First symptoms after sting	24 h	24 h	48 h	36 h
Duration of symptoms	10 months	9 months	8 months	10 months
Hymenopteran type	Honey bee	Honey bee	Honey bee	Common wasp
Positive RAST (CAP Pharmacia)	3.15 IU/mL	4.15 IU/mL	6.44 IU/mL	3.33 IU/mL
Total IgE	121 IU/mL	148 IU/mL	17.8 IU/mL	1625 IU/mL

sting; (b) none of the patients had a history of cold urticaria. Patient 3 reported a history of Hashimoto's disease and rheumatoid arthritis but reported no urticarial lesions in the past; (c) the relatively short duration of the clinical manifestations that lasted no more than 12 months. Provocation tests proved negative after 12 months; (d) the fact that laboratory investigation was within normal limits at the time of the hymenoptera sting and remained unremarkable during the following 12 months suggests there was no other triggering factor for the cold urticaria onset.

Pathogenesis of acquired cold urticaria is centred on mast cell activation. In some patients cold urticaria may be dependent on a transferrable IgE-like antibody. In cases of cryoglobulin detection it has been suggested that complement activation results in histamine secretion via anaphylatoxins.¹ Our patients showed neither cryoglobulin detection nor any indication of complement activation. These data are in accordance with those reported by Hogendijk and Hauser.² Pathogenetic mechanisms might prove to be a lot more complex. It is possible that platelet activation factors, neuropeptides and kinins are involved in mediator secretion by the mast cell.¹ Although specific sensitization to hymenoptera venom cannot be directly associated to acquired cold urticaria, one should bear in mind that sensitization to hymenoptera venom is high in Greece.⁶

It is our opinion that the reported cases of cold urticaria were acquired due to hymenoptera sting. We suggest that all patients presenting with this rare disorder should be questioned about recent hymenoptera stings when medical history is evaluated.

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Accepted for publication 23 June 2003

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Leucocytoclastic vasculitis and influenza immunization

We are very grateful to Dr Tavadia and colleagues for highlighting the link between leucocytoclastic vasculitis and influenza immunization.¹ We recently had cause to speculate whether influenza immunization had been responsible for a fulminant bullous necrotizing cutaneous vasculitis of the lower legs in a 64-year-old woman (Fig. 1) who had received inactivated influenza vaccine (split virion) 10 days earlier. Histology showed the changes of leucocytoclastic vasculitis. There was no evidence of systemic involvement. Despite prednisolone 40 mg daily her bullae progressed to deep ulcers which then took more than 3 months to heal.

Since such a large proportion of the elderly population in the UK receives influenza immunization, a temporal association between immunization and any acute illness is more likely than not to be coincidental. We were therefore reluctant to attribute our patient's vasculitis to this before we had excluded other causes. There were, however, no obvious alternative antecedent events to account for her vasculitis. Of the numerous oral medications she had been taking, only bendrofluazide, introduced 6 months earlier, was thought to be a possible aetiological factor. This was discontinued. A screen for other causes of vasculitis was negative. Dermatologists should be aware that influenza immunization must now be included in the large list of precipitants of cutaneous vasculitis.

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Figure 1 Bullous vasculitis on the lower limbs following influenza vaccination.

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Transverse leukonychia of the fingernail following proximal nail fold trauma

The presence of transverse leukonychia secondary to trauma is well reported;^{1,2} however, the time delay from injury to development of leukonychia is such that details of the original injury are often forgotten or are at best vague and rarely recorded. We report the first case of documented proximal nail fold trauma resulting in transverse leukonychia.

A 32-year-old man presented with a proximal nail fold haematoma of his left middle finger following DIY induced minor trauma (Fig. 1a). Following resolution of the haematoma no nail abnormality was detected until 2 months later, when transverse leukonychia was apparent (Fig. 1b).

Generalized transverse leukonychia is commonly a result of repeated trauma – particularly of the toes,² systemic illness,³ or systemic therapy⁴ – particularly chemotherapy.⁵ Isolated transverse leukonychia typically occurs following trauma to the nail matrix. The nail matrix extends proximally beneath the proximal nail fold and thus trauma in this region can lead to nail matrix abnormalities. In our case the trauma to the proximal nail fold demonstrates the link between damage to the nail matrix and the production of parakeratotic onychocytes, which retain their nuclei and reflect light leading to the development of leukonychia.

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Accepted for publication 12 September 2003

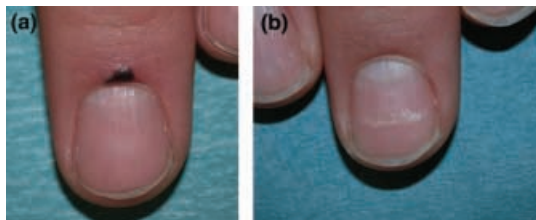


Figure 1 Left middle finger nail showing (a) proximal nail fold haematoma and (b) transverse leukonychia 2 months after initial trauma.

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Congenital dermoid fistula of the anterior chest region

We report a case of congenital dermoid fistula on the chest. A healthy 1-year-old Japanese boy was referred for evaluation of a congenital lesion on his anterior chest. The mother stated that the lesion had tended to swell and crust over for the previous 3 months, with recurrent infection and abscess formation. A local doctor had treated the lesion with oral antibiotics and incision and expression of contents, but without benefit.

On examination, a 2 × 1-cm soft, reddish, slightly elevated plaque with a crusted ulcer was present on the left anterior chest (Fig. 1). This lesion bled easily, and was tender but not purulent. On palpation a deeper component could be felt, and this was freely movable. There was no regional lymphadenopathy. Ultrasonography showed a 1 × 1-cm, cord-like low-density area. Skin biopsy demonstrated only granulation tissue with infiltration of polymorphonuclear cells. Culture grew nothing and mycobacterial infection was not detected from the skin lesion. A photograph that the mother had taken 6 months previously demonstrated a skin dimple at the same place as the lesion.

A diagnosis of congenital dermoid fistula was made. The fistula was excised completely under general anaesthesia. Histology showed a fistula lined by a keratinizing squamous epithelium with numerous hair follicles and sebaceous glands (Fig. 2). However, no eccrine or apocrine glands were seen. At 2-month follow-up, there was no recurrence of the lesion or development of new lesions. The family was pleased with the results.

This lesion is an uncommon congenital abnormality, but the clinical appearances are characteristic. Shuttleworth *et al.* used the term 'median nasal dermoid fistula' to describe this condition in the mid-line of the dorsum of the nose.¹ In Japan, Matsunaga *et al.* reported 18 cases of congenital fistula occurring in the anterior chest region, as in our patient.²



Figure 1 A soft, reddish, slightly elevated plaque, 2 × 1 cm, with a crusted ulcer on the anterior chest.

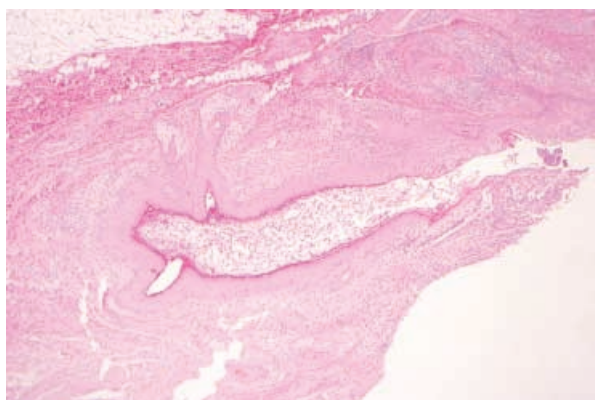


Figure 2 Fistula lined by a keratinizing squamous epithelium with hair follicles and sebaceous glands (haematoxylin & eosin).

If left untreated, a number of complications may occur, including abscess formation, and osteomyelitis of the underlying bone. Complete and adequate preoperative assessment and subsequent referral to a plastic surgeon are essential. Total excision of the fistulous tract is necessary for the optimal management of this condition.

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Accepted for publication 8 September 2003

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Narrow-band (TL-01) ultraviolet B phototherapy for chronic urticaria

Recent guidelines on the management of urticaria commented that the evidence for ultraviolet B (UVB) phototherapy was 'unconvincing'.¹ The report of a 15-patient open study suggested that broad-band UVB might be effective for chronic urticaria, particularly the physical urticarias,² but no controlled study of any form of UVB for urticaria has been conducted. Since 1985 we have used narrow-band (312 nm, TL-01) UVB as a treatment for chronic urticaria when simpler measures failed.

We present a retrospective review of the use of narrow-band (NB) UVB for chronic urticaria. Between 1986 and 2000, 94 patients (68 female) with chronic urticaria received a course of NB-UVB. Eighty-eight were recorded to have chronic 'idiopathic' urticaria. Their mean age was 44 years, ranging from 12 to 81 years, and most (88/94) were of skin phototypes I to III. The duration of urticaria prior to institution of NB-UVB varied from 3 months to 20 years. These patients had not responded to at least two H1 antihistamines, and most had experienced a variety of antihistamine combinations, and a trial of a diet low in salicylate and histamine releasing foodstuffs.

One hundred and thirteen NB-UVB treatment courses were administered (to these 94 patients) of which the records of 95 were detailed enough for evaluation. Patients were treated three times a week, with a starting dose 70% of minimal erythmal dose (just perceptible erythema at 24 h), and following a percentage incremental regimen adjusted according to erythema (and other, such as heat induced urticaria and polymorphic light eruption) responses. We aimed to follow our standard incremental protocols for psoriasis (lately 20% reducing to 10% increments), but our experience is that many patients with urticaria, as with atopic dermatitis, quickly move to a very much lower incremental regimen (such as 10% every third treatment). Anecdotally, we have an impression that near-erythmal doses of UVB can lower the threshold for dermographism, and that patients with a symptomatic dermographism component to their chronic urticaria require especially careful adjustment of doses.

Table 1 Outcome scoring scale for UVB treatment of chronic urticaria.

1	No change
2	Minimal improvement (no change in frequency or extent symptomatic improvement ±)
3	Moderate improvement (less frequent or extensive symptomatic improvement +)
4	Marked improvement (occasional episodes and less extensive symptomatic improvement ++ reduced antihistamine use)
5	Clearance with or (without antihistamines)

The median number of treatments was 22 (range, 2–37), and the median top dose was 1238 mJ/cm² (range, 100–2111 mJ/cm²). The immediate outcomes, documented at completion of each phototherapy course, recorded in the phototherapy unit were reviewed. Sixty-eight courses (72% of evaluable courses) produced clearance (40%), marked (15%) or moderate (45%) improvement, according to an outcome scoring scale (Table 1). Twenty-seven (28%) of courses were documented as unsuccessful. We were able to obtain follow-up data from clinic visits 4–12 weeks after course completion for 73 courses (for 63 patients). This selected group of patients were those judged to require a follow-up outpatient clinic appointment after completion of their phototherapy course. Fifty-eight courses (79%) produced a good response at follow-up, with clearance in 30%, marked improvement in 29% and moderate improvement in 20%.

In 2002 we attempted to contact the 94 patients who received NB-UVB for their chronic urticaria to find out if they felt that NB-UVB helped their condition, and if so, if the effect was sustained. Four patients had died, and of the remaining 90 we traced and conducted a telephone questionnaire with 57 (63%). Nineteen patients were clear (33%) and of the remaining 38 (66%) still suffering from chronic idiopathic urticaria, 26 (45%) felt that their condition had improved and only 12 (21%) felt that it was unchanged.

Chronic urticaria can severely reduce quality of life³ and can be difficult to control, sometimes necessitating therapies such as cyclosporin.⁴ If a simple, relatively safe (compared to systemic immunosuppressive therapies), and widely available treatment such as NB-UVB works in the majority of a group of patients unresponsive to first-line treatments, then this is an important approach.

A randomized controlled study to assess NB-UVB for the chronic urticarias is needed, and we are now in the process of designing such a trial. In the meantime, while awaiting good quality evidence of its value (or evidence that it does not really work), we regard NB-UVB as a useful second-line therapy for chronic urticaria.

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Accepted for publication 29 September 2003

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A further case of non-ILVEN

In a recent issue of this journal, Oskay and Kutluay describe a case of 'inflammatory linear verrucous epidermal naevus associated with ipsilateral undescended testicle'.¹ For the following reasons I should like to raise my voice of polite dissent.

Firstly, the clinical and histopathological photographs clearly show that this is an example of epidermal naevus of the epidermolytic type. Figure 2 presents neither parakeratosis nor acantholysis, but epidermolytic hyperkeratosis characterized by granular degeneration. Hence, this is a mosaic manifestation of epidermolytic hyperkeratosis of Brocq,² thus excluding a diagnosis of inflammatory linear verrucous epidermal naevus.

Secondly, the possibility that the epidermal naevus of this boy is causally related to his cryptorchidism can be excluded because otherwise male patients with epidermolytic hyperkeratosis of Brocq would often have undescended testicles. So far, such syndromic association has never been reported.

Moreover, Oskay and Kutluay state that 'ILVEN has been reported in association with ipsilateral soft tissue, skeletal and renal abnormalities'. In fact, such an association has never been described. All cases of so-called ILVEN associated with such extracutaneous defects represent examples of CHILD naevus, being the cutaneous hallmark of CHILD syndrome.³ No exception from this rule has so far been presented in the literature.⁴

In conclusion, this is a typical case of epidermal naevus of the epidermolytic type, and the ipsilateral cryptorchidism should be taken as a coincidental finding.

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Accepted for publication 15 October 2003

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