Ectopic Cilia in a Caucasian Girl with Atopic Eczema

Sir,

Ectopic cilia (EC) is a very rare condition, and only a few cases have been reported in the literature since the first description by Wiegmann in 1936 (1). We here present the case of a 12-year-old Caucasian girl with a history of atopic eczema and EC. To our knowledge, EC in a patient with atopic eczema has not yet been reported.

CASE REPORT

A 12-year-old Caucasian girl with atopic eczema according to the criteria of Hanifin & Rajka (2) was referred for evaluation of a congenital pigmented mole, measuring 1.8 x 1.2 cm, and extending horizontally in the upper inner quadrant of her right breast. Further dermatological examination disclosed a tuft of 10 dark, densely grouped cilia on the outer side of the girl’s right upper eyelid, 8 mm above the lash line (Fig. 1).

The aberrant cilia had been present at birth, and apart from the cosmetic aspect, caused no apparent inconvenience. There was no family history of any similar disorder. On palpation, the tuft appeared deeply rooted to the underlying tarsal plate, projecting freely through implanted lashes. As an extreme, Mackintosh & Grayson (9) found 2 cases of EC originating from the iris without any convincing history of injury to the eye, possibly due to lashes arising from a dermoid cyst or teratoma.

Fig. 1. A tuft of ectopic cilia, emanating 8 mm above the right lateral upper eyelid margin. Note also atopic eczema-associated features, including a poorly defined periorbital pink scaly rash, a vanishing of the lateral eyebrows (Hertoghe sign) and – indicated – extra infraorbital skin folds (Dennie-Morgan sign).

The aberrant cilia had been present at birth, and apart from the cosmetic aspect, caused no apparent inconvenience. There was no family history of any similar disorder. On palpation, the tuft appeared deeply rooted to the underlying tarsal plate, projecting freely through an aperture of the skin of the upper eyelid. Eversion of the right eyelid demonstrated a completely unaffected palpebral and bulbar conjunctiva. Macroscopically as well as microscopically, no difference in the morphology of cilia harvested from the tuft and the lash line could be found. Further ophthalmologic examination as well as general examination, including X-ray screenings and broad laboratory tests, were unremarkable. Elective excisional biopsy of the EC was refused.

The atopic eczema presented itself in a mild form (SCORAD-index (3) 13/103), where only adequate hydration of the skin with emollients and avoidance of irritants were necessary. The congenital mole was excised under local anaesthesia. Results of histopathologic examination confirmed the presence of a naevomelanocytic naevus. The patient’s subsequent postoperative course was uneventful.

DISCUSSION

The first report of EC was published in 1936 by Wiegmann (1). He described the case of a 5-year-old girl, in whom a curved cilium grew from the middle of the conjunctival surface of the right tarsus, in which it was deeply embedded.

In the majority of documented cases with this congenital abnormality, a cluster of cilia is found on the anterior surface of the eyelid (4–8). Reports on the degree of vertical separation of the ectopic cilia from the eyelid margin are varying. As an extreme, Mackintosh & Grayson (9) found 2 cases of EC originating from the iris without any convincing history of injury to the eye, possibly due to lashes arising from a dermoid cyst or teratoma.

According to the hypothesis of “Meibomian gland substitution” (10), the ectopic lashes originate from lash follicles, which completely or partially replace Meibomian glands. Bader (11) described a case with apparently normal eyelashes, in which some Meibomian glands were replaced by lash follicles, appearing as dark spots under the tarsal conjunctiva. Tavolara (12) reported a case in which two lashes originating in the tarsus grew into the conjunctival sac. Histological observations indicated the possibility of a substitution of some Meibomian glands by the abnormally implanted lashes.

In opposition to this hypothesis is the observation by Dalgleish (4), Owen (5) and Gordon et al. (8), who detected a normal Meibomian gland system in the cases where EC must be differentiated from abnormally placed hair. Dalgleish (4) emphasized the importance of histological examination of the lesion, with eyelashes characterized by the presence of sweat glands of the large apocrine type attached to the follicles. As in our case, where excision and histological examination were declined, most reports on this subject have failed to give proof of the presence of apocrine sweat glands attached to the hair follicles, which would verify the identity as cilia (1, 6, 7, 13, 14). Nevertheless, macroscopically as well as microscopically, no difference between cilia from the ectopic spot and the eyelid margin could be found in our patient.

In humans, the association of EC with a small number of abnormalities is documented. Distichiasis and EC is reported by Bader (11) and Tavolara (12). Gordon et al. (8) described a complex choristoma of the right eyelid, containing EC and functioning aberrant lacrimal gland tissue, intermittently producing tears.

As to the localized association of EC and cutaneous features of atopic eczema in the periorbital region in our case (Fig. 1), including a poorly defined scaly rash, a vanishing of the lateral eyebrows (Hertoghe sign) and extra infraorbital skin folds (Dennie-Morgan sign), no underlying common causative disorder became evident. Obviously, the observed association has to be evaluated as coincidental.

Treatment of EC consists of excision. Partial regrowth of cilia after excision at the original site is found (6), possibly due to an incomplete surgical procedure.

Palpebral EC is a very rare abnormality in humans, with only 11 cases documented in the literature. To our knowledge, this is the first reported case of such a lesion associated with atopic eczema.

REFERENCES

Malignant Skin Lesions on the Legs and Feet at a Dermatological Leg Ulcer Clinic during Five Years

Sir,

In clinical practice leg and foot ulcers are a common problem, with an increasing frequency in the elderly, as well as the number of skin cancers. It is important to have malignancies in mind both initially when the patient seeks help for a leg ulcer and also if the ulcer does not heal as anticipated. If a correct diagnosis is established at an early stage, the skin malignancy can be cured and spread stopped. The malignancy may be a primary skin cancer or a secondary malignant degeneration in a chronic skin lesion or ulcer (Marjolin’s ulcer) (1).

The aim of the present study was to estimate the occurrence of malignancies on the lower limbs and feet in the setting of a leg ulcer clinic and to see if there were any signs or demographic data that distinguished the malignant lesions from leg ulcers.

MATERIAL AND METHODS

Retrospectively from outpatients attending the leg ulcer clinic of the Dermatology Division of Sahlgrenska University Hospital, Göteborg, during a 5-year period (1991–1995) patients with biopsy-verified malignancy of the leg or foot were selected to be studied.

The following data were recorded at the time of biopsy verification: diagnosis, sex and age of the patients, size and type of skin changes (ulcerated or not), duration from the start of the lesion to diagnosis, and location. The number of patients that visited the clinic for non-malignant leg ulcers was registered.

Statistics

Values are given as mean, median and range (min-max) for different age groups. When comparing the age groups for malignant lesions and leg ulcers, the chi-square test with Yates (continuity) correction was used.

RESULTS

Six hundred and eighty-five patients (66% women, 449/685) had non-malignant types of leg ulcers and a mean age of 73 (median 75), range 17–99 years. The 20 patients (70% women, 14/20) with malignant lesions on their legs or feet had a mean age of 80 (median 80), range 58–92 years. Seventeen of these patients (71% women, 12/17) with a mean age of 82 (median 85), range 58–92 years, had basal and/or squamous cell carcinomas and these patients were significantly older than the patients with non-malignant leg ulcers (p < 0.00001).

There were more women than men among the patients with malignancies (ratio 2:3:1) than in the group of patients with non-malignant leg ulcers (1:9:1) (n.s.).

Malignant skin lesions were found in 3% (20 patients out of 705) of all patients at an ulcer clinic during 5 years. Three patients had less common types of malignancies. One patient had a cutaneous T-cell lymphoma, one patient a non-HIV-related Kaposi’s sarcoma and one patient a malignant melanoma on the leg.

The most common type of malignancy was basal cell carcinoma, found in 60% of the patients (12/20). Fifteen percent (3/20) of the patients had squamous cell carcinomas, 5% (1/20) had a base-squamous carcinoma and 5% (1/20) a carcinoma in situ. There were 29 malignant lesions in the 17 patients with the basal and squamous cell carcinomas. Treatment was mostly excision and grafting. The duration before diagnosis varied from 2 months to 3 years.

The site of the malignant lesions on the legs and feet had a large variation, but 67% (8/12) of the basal cell carcinomas were found on the anterior aspect of the leg.

In half of the cases (10/20) the skin lesions were ulcerated.

Four patients had simultaneously malignant lesions and non-malignant leg ulcers.

DISCUSSION

Primary or de novo malignant skin lesions in the lower limbs were claimed to be a very rare condition by Black in 1952 (2). Even rarer is a malignant transformation, a secondary malignancy, also called a Marjolin’s ulcer, in chronic venous ulceration (3). Transformation takes long, often 20–30 years, but at least 2–3 years (4). None of the malignant ulcers in this study was considered to be a secondary malignancy, since they had a relatively short duration, and there was no history of previous

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