Trichoepithelioma, Cystic Acne and 13-cis-Retinoic Acid

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Treatment with 13-cis-retinoic acid during 12 weeks of a patient with multiple trichoepitheliomas and acne cystica et comedonica did not affect the number of trichoepitheliomas. The cystic lesions, most of them trichoepitheliomas with only a few non-inflammatory cystic acne lesions, were not affected by this treatment. Key words: Trichoepithelioma; Acne cystica: 13-cis-Retinoic acid. (Received January 30, 1984.)

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Trichoepithelioma (epithelioma adenoides cysticum) is usually observed as multiple lesions, but is seen occasionally as a solitary tumor. Histologically different types of trichoepithelioma can be seen (1). The occurrence of multiple trichoepitheliomas is considered to be a benign autosomal dominant disorder, usually starting in adolescence, sometimes associated with cylindromas. Cylindromas have been reported to convert in malignant metastatic disease.

We report the result of treatment with 13-cis-retinoic acid (13-cis-RA) of a patient with multiple trichoepitheliomas on the face and acne cystica et comedonica in the neck.

CASE REPORT

A male patient, aged 41 years, was seen with multiple lesions on the face and in the neck form the age of 26. Incidentally lesions from both areas became inflamed. The family history did not reveal similar lesions on the face, but inflammatory lesions were said to occur in the neck of the father and an uncle of the patient. The patient wanted removal of his facial lesions.

Dermatological examination showed multiple translucent papules in the nasolabial folds and multiple elevated cystic lesions on the forehead, 0.3–1.5 cm in diameter. The neck showed about 50 black comedones and a few large cysts. Some inflammatory papules were seen in the neck and on the face.

Histological examination of biopsies of 3 cystic lesions on the forehead and 2 papular lesions in the nasobial folds showed multiple cysts with a fully keratinized centre aligned by several layers of basophilic cells. The size of the cysts was larger on the forehead than in the nasolabial folds. In the tissue surrounding the cysts strands and conglomerates of basophilic basaloid cell were seen. Two cystic lesions in the neck were biopsied. Both lesions contained a follicular epidermal cyst as can be seen in acne (2).

Treatment with 13-cis-RA, 1 mg/kg/day, was given during 12 weeks. The number of lesions (comedones, inflammatory papules, cysts and papular lesions in the nasolabial folds) was counted every two weeks. During the treatment period all comedones and inflammatory papules disappeared. The number of cystic lesions and lesions in the nasolabial folds (about 50) did not decrease. Before and during this treatment period transient inflammation of a few cystic lesions on the face and in the neck was seen. A control biopsy of two trichoepitheliomas after the treatment course showed no remarkable histologic changes.

Laboratory tests were performed regularly. Transient moderate elevation of alkaline phosphatase, serum glutamate pyruvate transaminase and triglycerides was noted. After the retinoid course the patient was treated with coagulation, excochleation and cryo-surgery with acceptable results.

DISCUSSION

Treatment of our patient with 13-cis-RA resulted in disappearance of the open comedones and inflammatory acne papules, but did not affect cystic lesions on the face and in the neck

and the papular trichoepitheliomas in the nasolabial folds. As indicated by the histology of 3 cysts on the face and 2 cysts in the neck, the majority of the cystic lesions on the face appeared to be trichoepitheliomas, whereas the few cysts in the neck appeared to be acne cysts. Clinically almost all cysts on the face and in the neck were non-inflammatory lesions

Biopsies of trichoepitheliomas before and after retinoid treatment did not reveal marked histologic changes due to therapy, cysts as well as basaloid strands and conglomerates still being present after the treatment course. This may accentuate the difference between genuine basocellular carcinoma cells and the basaloid cells seen in trichoepithelioma. In a study of patients with genuine basocellular carcinomas an effect of treatment with 13-cis-RA on the basocellular carcinomas was observed (3). The smaller carcinomas were reduced in size or disappeared altogether.

The absence of effect of 13-cis-RA treatment on the non-inflammatory cystic acne lesions in the neck of this patient confirms our observation in a study of 9 patients with acne conglobata, participating in a multicentered trial, that non-inflammatory cystic lesions in acne generally do not respond to treatment with 13-cis-RA (4).

We conclude that 13-cis-RA seems to be of no value in the treatment of trichoepitheliomas.

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