Isotretinoin for Sebaceous Skin Lesions in Muir-Torre Syndrome: A Case Report

Sir,
Sixteen years ago, a 55-year-old man underwent removal of low-to-average differentiated adenocarcinoma, penetrating the walls of the colon, at which time metastases were found in three abdominal lymphoid glands. During the following years, he was examined regularly, had repeated coloscopies and/or rectoscopies and required extirpation of several rectal and sigmoidal polyps, that mostly showed a tubulovillous adenoma, with slight dysplasia on histological examination. Additional resections of the colon and rectum were also done. His mother had had a brain tumour and his father died of lung cancer, but neither had had facial lesions.

CUTANEOUS HISTORY
At the age of 44, he developed erythematous lesions and telangiectasias on the tip of his nose. In addition, scattered non-pruritic, yellowish papules and nodules (the largest two being 12 mm in diameter) were found on his cheeks, forehead and nose (Fig. 1). The clinical diagnosis was sebaceous adenoma, but a biopsy from a papule on the forehead showed sebaceous hyperplasia. The lesions were removed with a razor, but they recurred rapidly. Isotretinoin [0.5 mg/kg/day (weight 80 kg)] was given for 15 months. After 6 weeks of treatment, nearly all the sebaceous hyperplasia lesions and nodules had flattened and resolved (Fig. 2). After stopping the treatment for 1 month, the lesions recurred, but they disappeared promptly, as before, when isotretinoin was given again (0.85 mg/kg/day). After 14 months, the therapy ceased and the lesions became visible again within 2 weeks. After vaporization with CO₂ laser down to the level of the surrounding skin, there was a rapid recurrence. Isotretinoin was re instituted, as before, for 9 months, with good effect. The sebaceous lesions that rapidly recurred after 1 month were again removed by laser. During the following years, the sebaceous lesions on his face were aggressively treated once a month with CO₂ laser. Nevertheless, the lesions reappeared quickly. The surgical measures were therefore abandoned and isotretinoin was given again, with a quick response, but the skin lesions recurred when the medication was stopped. Four years ago, he suddenly developed a fast-growing tumour on his left lower arm. Excision and histological examination showed cancer, originating in the sebaceous glands. For 4 years past he has been continuously given isotretinoin, in doses varying between 0.38 and 0.75 mg/kg/day, with satisfactory control.

During treatment, the laboratory data were regularly checked and the levels of alanine aminotransferase, alkaline phosphatase and γ-glutamyltransferase were elevated. The triglyceride levels rose to the upper limit of the reference range (< 2.2 mmol/l) and sometimes to more than twice the upper limit, but never above 10 mmol/l. The cholesterol levels were always normal. He also had a constantly high leukocyte count.

COMMENT
To diagnose the Muir-Torre syndrome, sebaceous adenoma, sebaceous epithelioma or sebaceous carcinoma, and even keratoacanthoma, should be present together with a tumour in the intestine (1). This patient developed a skin cancer derived from sebaceous cells after 10 years of observation and he now fulfills the criteria for the Muir-Torre syndrome (1–3). Specific sebac-
eous tumours can start before or at the same time as the visceral tumour or after it (1, 2).

Our patient benefited from the isotretinoin therapy, which has proved effective in managing many disfiguring sebaceous lesions. In two other cases of Muir-Torre syndrome, total or nearly total resolution of hypertrophic sebaceous glands was reported following isotretinoin treatment, and no new lesions were detected during the 10 to 14 month follow-up period without this medication (4). This contrasts with our experience, since the sebaceous lesions recurred within 1 month of discontinuation of isotretinoin. However, in accord with our findings, a case of “premature sebaceous gland hyperplasia” in a 37-year-old woman also showed a dramatic response of the lesions to treatment with isotretinoin (5), but the lesions recurred.

REFERENCES

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