# Development and Progression of a Periorbital Sebaceous Gland Carcinoma *In situ*

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Sebaceous carcinoma (SC) is a malignant neoplasm originating from sebaceous gland cells and occurring in 75% of cases in the periorbital region (1). Until now SC has been thought to arise *de novo* or sporadically out of a sebaceous naevus (2). We present here the first case in which a benign sebaceous adenoma developed into a premalignant SC *in situ*. Furthermore, in our second case a SC *in situ* and a SC were seen simultaneously in the histopathological excision slide of one tumour, suggesting that a SC *in situ* can break through the boundaries of the basal membrane and develop into a SC with metastatic potential.

## CASE REPORTS

### *Case 1*

An 82-year-old woman presented to our dermatology outpatient clinic with a 4-year history of an asymptomatic papule on the left eyebrow. Clinical examination showed a 7 mm diameter yellowish papule, with a clinical differential diagnosis of a hyperplastic sebaceous gland, xanthelasma or a basal cell carcinoma (BCC) (Fig. 1a). A 3 mm biopsy was taken, showing a sebaceous adenoma characterized by a sharply circumscribed proliferation of enlarged sebaceous lobules, composed of central fully mature sebocytes, and peripherally-disposed basaloid epithelial cells. A fibrous stroma separated these lobules from the adjacent dermis (Fig. 1b). Because of the benign character of this asymptomatic lesion, no further intervention was performed. Over a period of 6 years the lesion grew slowly and a further punch biopsy was taken. Again, a proliferation of enlarged sebaceous lobules was seen, completely surrounded by a basal membrane. This process was characterized by atypical cells with large polymorphic nucleoli and cells with clear cytoplasm and sebaceous differentiation; mitotic figures were frequent (Fig. 1c). This tumour, diagnosed as a SC *in situ*, was excised completely with a 5 mm margin. After one year of clinical follow-up, the patient remains free of clinical recurrence.

### *Case 2*

An 82-year-old woman was treated with complete surgical excision for a histologically reported nodular BCC on the right lower eyelid. At a follow-up visit 2 years later a clinical recurrence was suspected. However, a biopsy on this occasion was reported as Bowen's disease and the lesion was treated with cryosurgery. Six years after the initial surgical excision, the patient was referred to the Maastricht University Medical Centre because of progression of the lesion, despite multiple sessions of cryosurgery. On examination we noted a 15 mm erythematous shiny telangiectatic plaque and excised the lesion with a 3 mm margin (Fig. 2a). Histopathological examination showed a SC *in situ*, with tumour cells extending to the left lateral border of the histological margin. The original excision slide, which was initially diagnosed as a nodular BCC, was reviewed by our dermatopathologists and determined as a SC *in situ* with dermal invasion, and thus was diagnosed as a SC with metastatic potential (Fig. 2b). Mohs' micrographic surgery (MMS) was suggested by the multidisciplinary head and neck oncology team, but the patient refused further intervention.

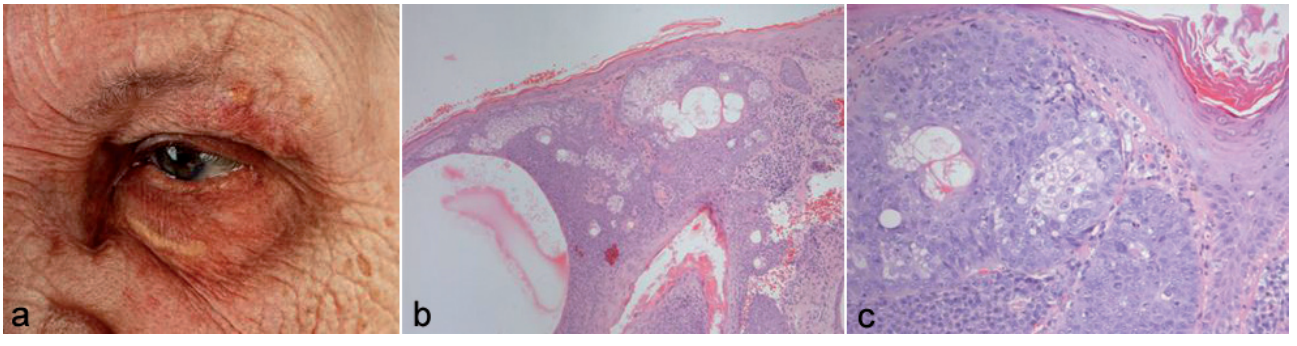


*Fig. 1.* Case 1. (a) Clinical presentation showing a yellowish papule of 7 mm on the left lateral eyebrow. (b) Sebaceous adenoma showing a sharply circumscribed proliferation of enlarged sebaceous lobules, composed of central fully mature sebocytes, and peripherally-disposed basaloid epithelial cells. Haematoxylin and eosin (H&E) original magnification ×100. (c) Sebaceous carcinoma *in situ* characterized by a proliferation of enlarged sebaceous lobules consisting of atypical cells with large polymorphic nucleoli and cells with clear cytoplasm with sebaceous differentiation. Mitotic figures are frequent. H&E original magnification ×200.

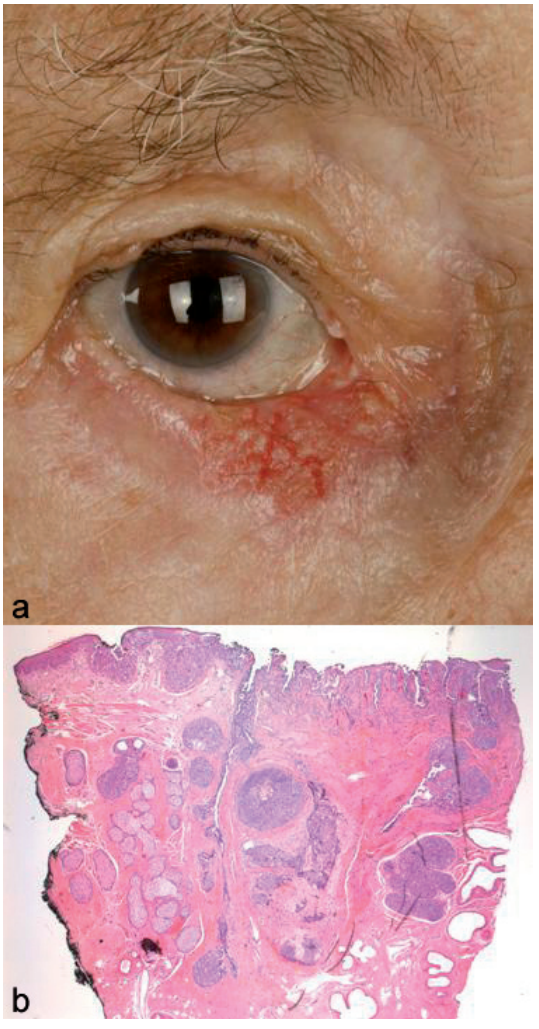


*Fig. 2.* Case 2 (a) Clinical presentation showing an erythematous shiny plaque of 15 mm with telangiectasias on the right lower eyelid. (b) Sebaceous carcinoma characterized by nests of atypical high mitotic cells with focally a basaloid aspect, foamy sebaceous gland cells and central necrosis, growing around the skin appendages within the basal membrane but locally suspect for invasive growth. Haematoxylin and eosin (H&E) original magnification ×25.

## DISCUSSION

Little is known about the incidence and aetiology of SC *in situ*. It is believed to be a rare premalignant stage of a SC without metastatic potential. The biological behaviour of development into an invasive carcinoma is unknown (3). To our knowledge, our second case presents for the first time the development of a SC *in situ* into a SC. The typical clinical presentation of a SC *in situ* is an erythematous or yellowish nodule or plaque with ulceration or crust formation. Clinically and histopathologically SC *in situ* is frequently misdiagnosed. Confusion with other epithelial (pre)malignant tumours of the skin is common, as seen in our second case. A correct diagnosis is essential because SC is an aggressive tumour, and delay in treatment can have a negative effect on prognosis. The diagnosis of a SC *in situ* can only be made histopathologically, preferably by a wedge excision. Microscopically, SC tends to extend beyond its clinical margins (4). Therefore the treatment of choice for SC remains complete surgical excision with 5 mm margins or MMS (1, 5, 6). The optimal treatment for SC *in situ* is controversial, because its natural history is unknown. Based on our second case, and the fact that SC *in situ* can grow deep into the dermis along the skin appendages, cryotherapy seems to be insufficient. We therefore recommend complete surgical excision.

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