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Sir, Poorly differentiated adenocarcinoma positive for gross cystic disease fluid protein-15 in the lacrimal drainage system: a case report and its implications for tumour origin

Primary poorly differentiated adenocarcinoma in the ocular region with positive immunoreactivity for gross cystic disease fluid protein-15 (GCDFP-15), a marker for apocrine epithelium, is a rare neoplasm with only a few reported cases.^{1–3} This neoplasm is considered to arise from the accessory lacrimal glands and caruncle as well as Moll's glands.³

Case report

A 79-year-old man noticed swelling of his right lower eyelid gradually worsening for 1 month (Figure 1a). Computed tomography revealed a solid tumour in the lower eyelid and medial and inferior orbit (Figure 1b and c). The biopsy specimen showed polygonal cancer cells with prominent nucleoli and granular eosinophilic cytoplasm proliferating diffusely and forming a pseudoglandular structure (Figure 2a and b). Immunohistochemically, these cancer cells were positive for cytokeratins (AE1/AE3) and GCDFP-15, and negative for S100, HMB45 and CD31/34 (Figure 2c and data not shown). Systemic examinations revealed no tumorous lesions in any organ other than the eyelid/orbit.

The patient underwent an exenteration of the orbital contents. The specimens showed extensive infiltration of cancer cells in the subcutaneous tissue of the eyelid/orbit and the lacrimal drainage system. However, no tumour involvement of the Moll's glands or caruncle was observed. To examine the tumour origin, we next performed immunohistochemical analysis of GCDFP-15 in the ocular region. As shown in Figure 2d–f, the non-goblet epithelial cells in the lacrimal sac were positive for GCDFP-15, as they were in the Moll's glands.

Comment

We describe a case of primary poorly differentiated adenocarcinoma in the ocular region that showed intense immunoreactivity for GCDFP-15, but did not involve the Moll's glands or caruncle. Shintaku *et al*² reported a case of GCDFP-15-positive ocular tumour, which, as in this case, was located around the lacrimal drainage system, suggesting the possibility that the tumours originated from this region.

GCDFP-15 is an excellent tissue marker for the apocrine epithelium, whereas some non-apocrine tissues, such as serous cells in salivary glands, also show immunostaining with GCDFP-15.⁴ Here, we revealed for the first time that non-goblet epithelial cells in the lacrimal sac are also immunopositive for GCDFP-15. Together, these findings suggest that the GCDFP-15-positive cells in the lacrimal sac could be the origin of this neoplasm.



Figure 1 Initial clinical presentation. (a) The right palpebral fissure is narrowed because of the swelling of the right lower eyelid. Palpation revealed a subcutaneous mass in the lower eyelid and medial orbit (arrows). (b and c) Axial (b) and coronal (c) computed tomograms showing a solid tumour extending into the lower eyelid and the medial and inferior portions of the orbit (arrows).

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Figure 2 Tumour histopathology and GCDFP-15 expression in the lacrimal sac. (a) The striated muscular bundles in the right lower eyelid were infiltrated diffusely by round or polygonal tumour cells. The tumour cells had large, atypical nuclei with prominent nucleoli and abundant eosinophilic cytoplasm. (b) The tumour cells focally formed a pseudoglandular structure. (c) The tumour cells showed intense immunostaining with GCDFP-15. (d–f) Analysis of GCDFP-15 expression in the eyelid and lacrimal drainage system. The non-goblet epithelial cells in the lacrimal sac showed strong immunostaining with GCDFP-15 (d) but were negative for control IgG antibodies (data not shown). (e) High-powered view of the surface epithelium of the lacrimal sac. (f) Immunoreactivity for GCDFP-15 in the Moll's glands of the lid margin.

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