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CASE REPORT

Further evidence for adipocytic differentiation by the neoplastic myoepithelium

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Lipomatous pleomorphic adenoma (PA)/myoepithelioma is rare. Nevertheless, adipocytic differentiation in salivary gland tumors is a well-known phenomenon. As extensive lipometaplasia occurs primarily in adenomas with myoepithelial participation, circumstantial evidence implicates neoplastic myoepithelium (NME), especially the spindle type, as a key partaker. We report here a unique PA that represents an equivocal transition from the epithelioid NME to adipocytes in recurrent lesions but not in a primary tumor.
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Case report

A 32-year-old woman noticed two small masses in the submandibular soft tissue. About 17 years before, pleomorphic adenoma (PA) of the left submandibular gland without adipose content had been removed in another institution (Fig. 1a). Recurrent lesions were composed of scar tissue and contained multiple encapsulated nodules of PA rich in epithelioid neoplastic myoepithelium (NME) including plasmacytoid cells. Neither myxoid nor chondroid element was present. Adipocytic changes started selectively inside the solid nests with little intervening stroma. Variably vacuolated lipoblast-like cells coalesced to form clusters of univacuolar adipocytes, resulting in a vague lobular arrangement of fully evolved fat (Fig. 1b–d).

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Epithelioid tumor cells were immunopositive for AE1/AE3, MNF116 and 34βE12 (Fig. 2a), but none expressed CAM5.2, cytokeratins 7 and 20, and epithelial membrane antigen. The staining for vimentin (Fig. 2b), glial fibrillary acidic protein (Fig. 2c) and S-100 protein (Fig. 2d) was strong and diffuse. Overall profile was typical of NME (1–3). A majority of multilocular adipocytes also exhibited a similar immunophenotype. Interestingly, NME expression of α-smooth muscle actin and calponin was completely negative in lipomatous areas (Fig. 2e), but intensely positive in the nearby non-lipomatous areas (Fig. 2f).

Comments

There is concern that NME is programmed to undergo bidirectional lines of differentiation, displaying both epithelial and mesenchymal tissue types (1). In the most common scenario, a variety of tumor-related stroma in PA including hyaline, myxoid and chondroid matrices are the products of NME (3). On the other hand, the origin of a prominent and extensive adipose content in PA and/or myoepithelial tumor (MET) has been a source of major debate. The current concept is that adipose tissue can be a consequence of lipogenic modification of spindle-shaped NME. Ideally, a transition pathway from NME through multilocular adipocytes to finally large univacuolar fat cells should be identified, but this sequential event has been only very rarely demonstrated. Unlike lipomatous PA/MET composed principally of spindled NME (4–6), the primary proliferating cells in our tumor were epithelioid in appearance. Given the morphologic and functional plasticity (1), other than spindle type, epithelioid NME may also be responsible for adipocytic differentiation. Although experience is limited, the absence of myogenic markers in lipomatous areas offers alternative hypothesis that epithelioid NME rarely modify its matrix producing ability along with lipogenic line, unless its indigenous smooth muscle property is lost. The molecular mechanism governing diverse phenotypic modifications by NME is worthy of further study.

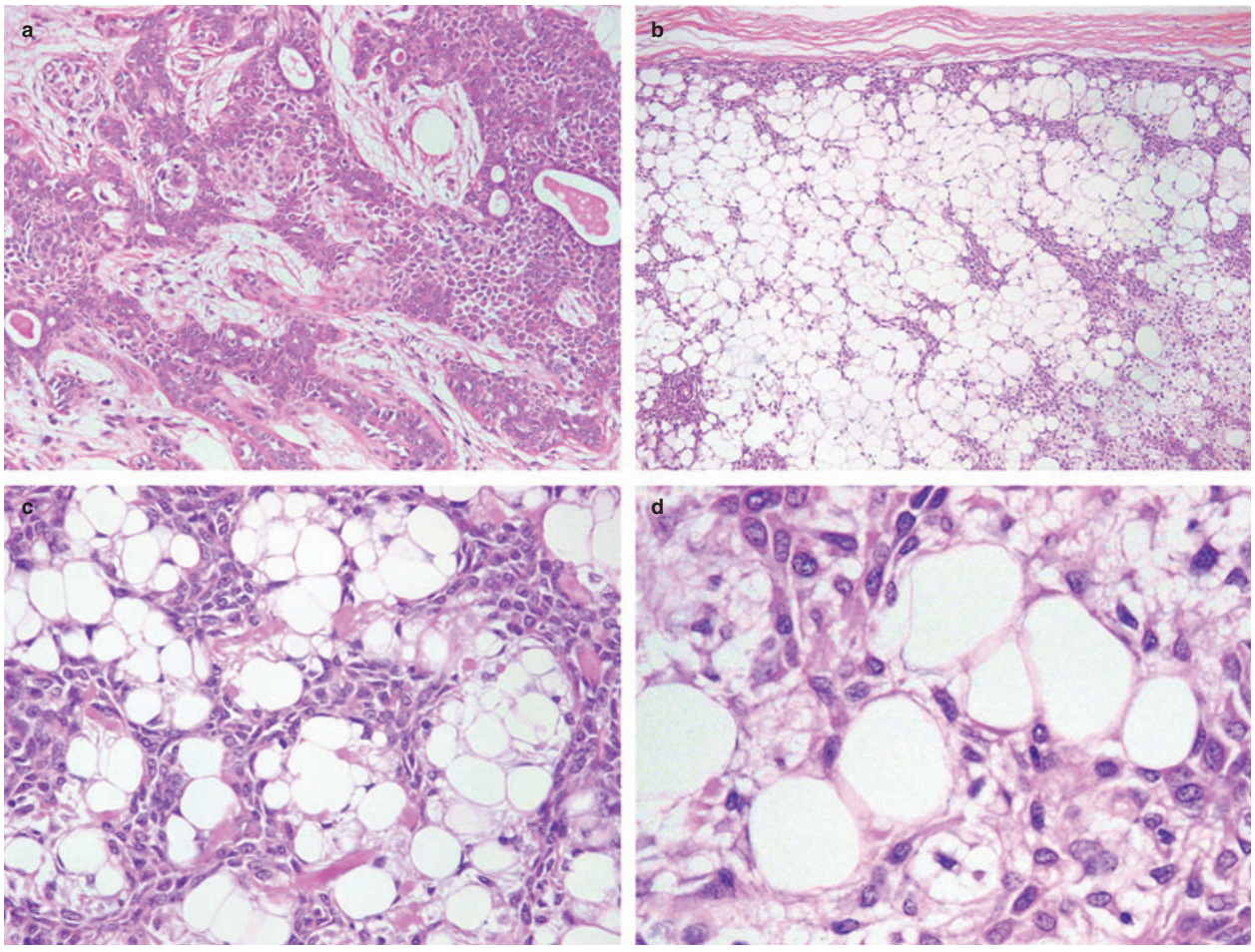

Figure 1 (a) Primary tumor lacks adipose tissue (H&E, ×200). (b–d) Epithelioid and adipocytic cells imperceptibly blend with each other forming a continuum with intermediate types in a recurrent lesion (H&E, b × 100; c × 400; d × 600).

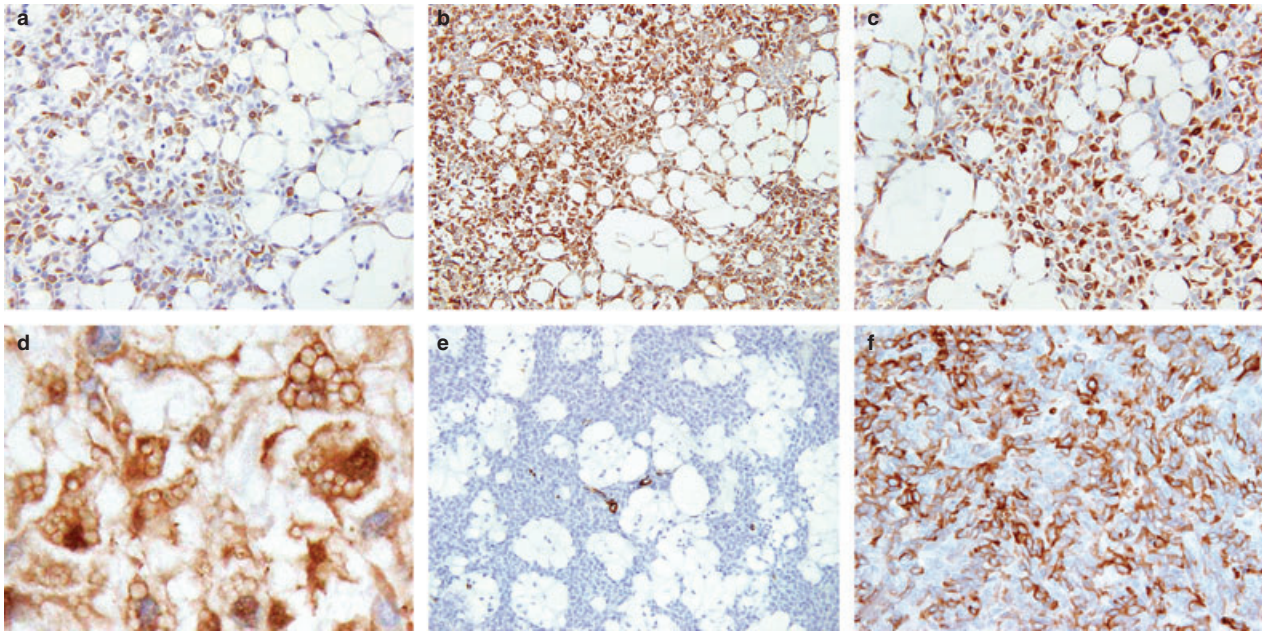

Figure 2 Epithelioid myoepithelial cells and adipocytes express for AE1/AE3 (a), vimentin (b), and glial fibrillary acidic protein (c). Multilocular configuration of lipoblast-like cells is highlighted by S-100 protein (d). Myoepithelial expression of α-smooth muscle actin is absent in lipomatous area (e) but detectable in non-lipomatous area (f). (ABC method, a, b and e × 200; c and f ×400; d, ×600).

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