Letter to Editor
A pelvic cellular solitary fibrous tumor with multifocal expression of cytokeratin AE1/AE3

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We read with great interest a recently published article by Lecoutere et al [1] entitled “multifocal cytokeratin expression in pleural and abdominal malignant solitary fibrous tumor: an unusual diagnostic pitfall”. Authors showed three histologically and immunohistochemically well-documented cases of pleural and abdominal malignant solitary fibrous tumor (SFT) with unexpected multifocal expression of cytokeratin AE1/AE3, which can cause significant diagnostic confusions for practical pathologists.

We have recently encountered a case of cellular SFT of pelvic cavity which showed multifocal immunoreaction to AE1/AE3, an unexpected phenomenon for STF which could elicit a broad spectrum of differential diagnostic considerations in this clinical setting. The patient was a previously healthy 53-year-old woman, who was incidentally found to have a pelvic mass measuring 9-cm in greatest diameter by annual physical examination. Tumorectomy was proceeded. Histological examination showed a well-demarcated, encapsulated tumor with a tightly packed, patternless, occasionally hemangiopericytic arrangement of cytologically bland, fusiform to ovoid cells with indistinctive cytoplasmic borders, setting in a fibro-collagenous stroma (Figure 1A and 1B). Mitotic activity was 4/50 high power filed, foci of necrosis were not noted. By immunohistochemistry, the tumor cells showed diffuse and strong positivity for Vimentin, CD34 (Figure 2), and bcl-2, as well as strong and patchy positivity for cytokeratin AE1/AE3 (Figure 3A and 3B), but no expression of smooth muscle actin, desmin, estrogen receptor, progesterone receptor, CD10, EMA, cytokeratin 5/6, cytokeratin 7, calretinin, D2-40, inhibin, or S100 protein. Ki67 stain decorated less than 5% tumor cells.

On the basis of the convincing morphologic appearances, together with the immunohistochemical profiles of CD34+, bcl-2+, and vimentin+, a diagnosis of cellular SFT of the pelvic cavity was rendered. With regard to the differential diagnoses of this tumor, the unforeseen strong positivity for cytokeratin AE1/AE3, despite multifocal, together with the clinical setting of a pelvic tumor may point to sarcomatoid carcinoma, arising either from the urinary tracts or from the genital organs, sarcomatoid mesothelioma originating from the peritoneum, and synovial sarcoma as major differential diagnoses. However, absence of histologically malignant features and no expression of other epithelial markers (EMA, cytokeratin 5/6, cytokeratin 7) and mesothelial markers (D2-40, calretinin, cytokeratin 5/6) in this tumor could readily excluded the possibility of a carcinoma or a mesothelioma. Synovial sarcoma often expressed vimentin and bcl-2, however, it usually lacks the expression of CD34 [2].

Aberrant cytokeratin expression by fibroblastic/myofibroblastic soft tissue lesions is not uncommon, such as inflammatory myofibroblastic tumors [3], proliferative fasciitis [4], and so on. Anomalous cytokeratin expression by SFT, usually limited and focal, has also been reported sporadically in the literature, most of which are documented in malignant cases [1, 5, 6]. Most recently, in a study by Barak et al [4] who investigated the immunoreactivity for calretinin and cytokeratin in a large cohort of myofibroblastic tumors, 3 out of 27 (11%) SFTs showed focal positivity for cytokeratin AE1/AE3 and cytokeratin 18. For practical pathologists, increased awareness of such nonclassical immunophenotype in SFT, together with the application of ancillary studies to exclude other differential diagnoses is crucial in arriving at accurate diag-
Cellular SFT with expression of CKAE1/AE3

Figure 1. Morphology appearance of a patternless, hemangiopericytic arrangement of tightly packed, cytologically bland tumor cells (A. H&E × 100; B. H&E × 200).

Figure 2. Diffuse strong expression for CD34 (CD34 × 200).

Figure 3. Multifocal strong cytoplasmic immunoreactivity for cytokeratin AE1/AE3 (A and B. Cytokeratin AE1/AE3 × 200).

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Disclosure of conflict of interest

None.