were surface microvilli, abundant intracellular filaments, desmosomes and intercellular spaces.

Immunostaining for Factor VIII related antigen, using a conventional immunoperoxidase bridge technique on formalin-fixed material, was negative in all areas of the tumour.

Discussion

Small adenomatoid tumours do occur in the uterus (Tilman 1980), but these never measure more than 2–3 cm in diameter, and this is the first report of a giant cystic adenomatoid tumour in this location. This variant of adenomatoid tumour is very rare. The majority have occurred in the ovary (Jones & Donovan 1965) and one case in the epididymis (Fajers 1949).

The histogenesis of adenomatoid tumours is still in doubt, but evidence based on transmission electron microscopy, scanning electron microscopy (Morris, Staff & Oates 1986) and immunohistology (Said, Hash & Lee 1982) supports a mesothelial origin. The occurrence in this tumour of areas of typical papillary mesothelioma as well as areas typical of adenomatoid tumour provides further support for this view.

References


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Brief report

Cytokeratins in smooth muscle cells and smooth muscle tumours

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A keratin positive metastatic leiomyosarcoma in the lung, which resulted in diagnostic error, is reported. The results of additional studies of 17 benign and malignant leiomyogenic tumours with various keratin antibodies are presented and discussed in the light of recent bibliographical data.

Keywords: cytokeratin, leiomyoma, leiomyosarcoma, metastasis

We have recently observed positive staining for epithelial markers in a metastatic leiomyosarcoma which led to an evident diagnostic error. It is significant to note that metastatic tumours were not analysed in the previous studies on this subject (Brown et al. 1987, Norton, Thomas & Isaacson 1987).
Case report

In September 1986 we first reviewed a bronchial biopsy of a man aged 55 years with a tumour in the upper lobe of the right lung. On light microscopy the bronchial wall was densely infiltrated by relatively large, polygonal malignant cells which were difficult to classify further (Figure 1). We found evident diffuse cytoplasmic positivity with the monoclonal cytokeratin antibody CAM 5.2 and with a polyclonal rabbit anti-keratin serum. Based on these results a primary bronchial, or metastatic poorly differentiated carcinoma was diagnosed. Two weeks later the patient was admitted to hospital with intestinal bleeding. At laparotomy an ulcerated 10 cm tumour was removed from the small bowel. The light microscopical appearances were consistent with a leiomyosarcoma: this was further confirmed by electron microscopy. Immunohistochemical studies showed not only vimentin, actin and desmin positivity, but surprisingly also a reaction with the polyclonal rabbit anti-cytokeratin serum and CAM 5.2 (Figure 2). Staining with the broadly cross-reacting monoclonal cytokeratin antibody RCK 102 was negative. In view of these findings the pulmonary lesion was accepted as a metastasis of the proven intestinal leiomyosarcoma. No other primary has been found and the patient is free of symptoms 10 months after operation.

Additional studies

In an attempt to explain the unusual staining pattern of a smooth muscle neoplasm, we tested 17 randomly chosen smooth muscle tumours as well as several normal myometrial excisions (frozen sections) from our files using the polyclonal rabbit anti-keratin serum (K40) and two monoclonal antibodies to keratins, i.e. CAM 5.2 and RCK 102 (a broadly cross-reacting antibody recognizing keratins 5 and 8). In all tumour cases formalin-fixed and paraffin-embedded material was available and in four cases additional fresh frozen material could be used. We also noted the staining patterns of the normal myometrium surrounding the tumours. The results are shown in Table 1. All the tumours except the leiomyoblastoma showed positivity for vimentin, actin and desmin. The positivity and intensity of staining for the kera-

Figure 1. Biopsy of the bronchial wall showing a dense infiltration of malignant cells, with strong cytoplasmic staining for CAM 5.2. Note the strong positivity of the superficial respiratory epithelium. ×120. Inset. ×300.
Cytokeratin expression in smooth muscle and smooth muscle tumours

<table>
<thead>
<tr>
<th>Material</th>
<th>No. of cases with positivity after incubation with following antisera</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal myometrium (7)</td>
<td>CAM 5.2</td>
</tr>
<tr>
<td>Leiomysomas of uterus (8)</td>
<td>3</td>
</tr>
<tr>
<td>Leiomyoablastomas of uterus (3)</td>
<td>1</td>
</tr>
<tr>
<td>Leiomysarcomas (8) (6 in uterus, 2 in ileum*)</td>
<td>5</td>
</tr>
</tbody>
</table>

*Includes present case report.

Discussion

Our observations, therefore, confirm the results of Norton et al. (1987) and Brown et al. (1987), but are contradictory to the results of Leader et al. (1986) who found negativity with CAM 5.2 antiserum in 22 leiomyosarcomas. The fact that we observed positive staining in smooth muscle cells and leiomyogenic tumours not only with CAM 5.2 antiserum but also with a polyclonal keratin antiserum and the monoclonal antibody RCK 102, indicates that it is becoming more and more plausible that keratins may indeed be present in smooth muscle cells. The reaction of smooth muscle cells with six completely different monoclonal antibodies (LE 61, LP 34, KL 1, CAM 5.2, RCK 102 and RCK 105) and one rabbit anti-keratin serum strongly supports this.
assumption. A final explanation of this phenomenon will, however, have to come from immunoblotting studies which, so far, have not given conclusive results in our laboratory.

Further support for our results can be found in the recent study of Van Muijen, Ruiter & Warnaar (1987) who, using anti-cytokeratin antibody M 20, observed positive staining in human fetal myocardium and adult myometrium indicating the presence of cytokeratin 8 in these tissues. The reaction with the antiserum LP2K and the results of two-dimensional gel electrophoresis indicated the presence of cytokeratin 18 and 19 (Van Muijen et al. 1987).

References


