Virchows Arch (2002) 440:105–106
DOI 10.1007/s00428-001-0546-0

LETTER TO THE EDITOR

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Pilocytic astrocytoma arising in a dermoid cyst of the ovary: a case presentation

Received: 9 August 2001 / Accepted: 5 September 2001 / Published online: 27 October 2001

Sir,

We present an exceptionally rare case of pilocytic astrocytoma in an ovarian dermoid cyst (mature cystic teratoma), occurring in a nulliparous 22-year-old woman. She presented with the chief complaint of lower abdominal pain and vomiting of 3 days duration. Menses were regular. Physical examination revealed a left-sided, abdominal, non-tender, movable, cystic mass. The external genitalia were normal. Sonography showed a left-sided multilocular mass, measuring 9 cm in its greatest diameter. The right ovary appeared normal. No ascites were seen.

At surgery, a cyst with a smooth surface emerging from the left ovary was removed by means of unilateral salpingo-oophorectomy. The capsule remained intact. No other tumorous lesions were palpated. The cut surface of the cyst which measured 6,5×4,5 cm revealed a "dermoid cyst", filled with yellow fatty material and hairs, while, within the cystic wall, small foci of cartilage and bone formation were noticed. On microscopic examination, the cyst was lined with epidermis with skin appendages. The cyst wall contained mature lipomatous, cartilaginous, and bone tissues. In a more solid area of the cyst, centrally located, glial tissue was noticed. The latter was sharply demarcated from the adjacent fibrofatty tissue (Fig. 1a) and showed transformation to pilocytic astrocytoma. This focus measured 0.8 cm at its greatest diameter. It was formed of loosely knit tissue composed of stellate and/or elongated astrocytes in microcystic regions (Fig. 1a, b). Rosenthal fibers, better appreciated as bright-red sausage- or corkscrew-shaped bodies on Masson trichrome stained sections, were obvious throughout the lesion (Fig. 2a). Mitoses were very rare. The tumor showed strong immunoreactivity for glial fibrillary acidic protein (Fig. 2b). The histological diagnosis was "mature cystic teratoma with a focus of pilocytic astrocytoma (grade-I astrocytoma, World Health Organization

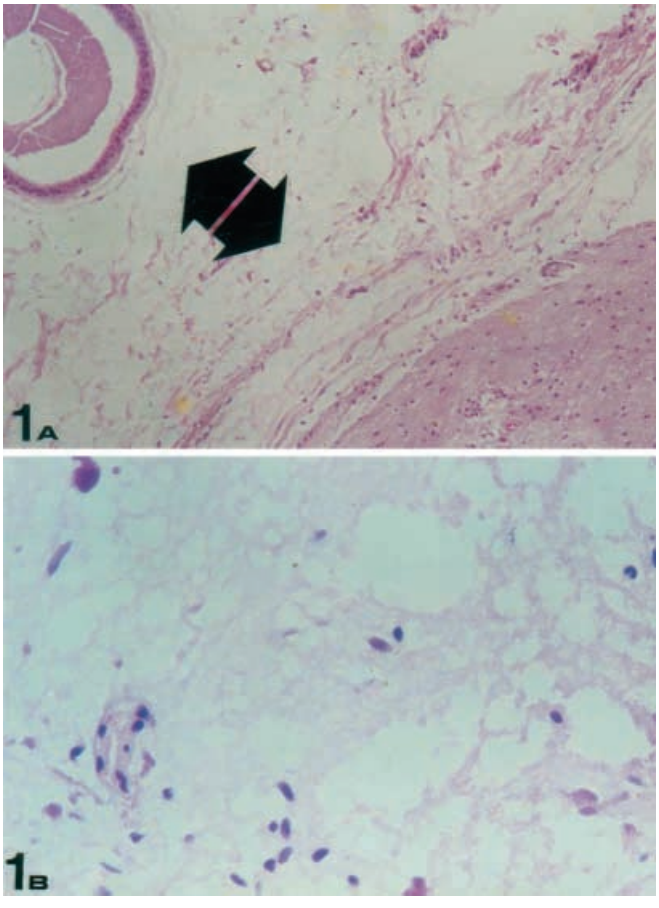

Fig. 1 **a** Sharp demarcation of pilocytic astrocytoma (*bottom left*) from the adjacent fibrofatty tissue. A hair follicle is well discerned at the *upper right* (hematoxylin and eosin ×120). **b** Microcystic pattern of the tumor in this area (hematoxylin and eosin ×340)

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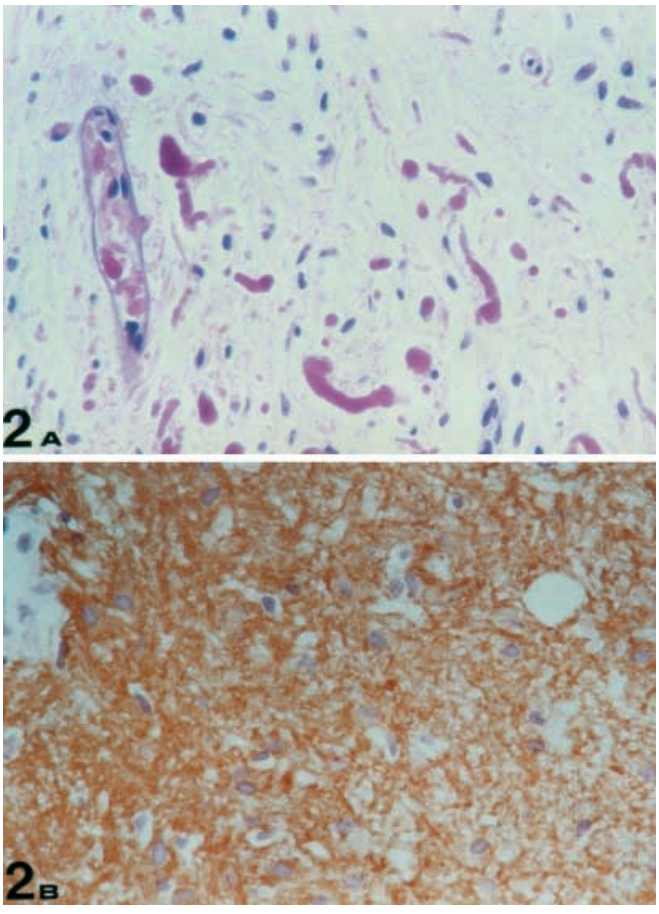

Fig. 2 **a** Pilocytic astrocytomas made up from rounded and spindle cells within a knit stroma. Rosenthal fibers are obviously bright red with Masson trichrome stain (×340). **b** Glial fibrillary acidic protein expression by the cells and neuropil (×340)

1993 classification)". Since the tumor was completely restricted within the dermoid cyst, which had been completely removed, no adjuvant therapy was given. Today, after 8 months follow-up, the patient is well, without evidence of disease.

Nervous tissue is present in at least four-fifths of all teratomas [1, 2]. Reports on astrocytic tumors arising in mature cystic teratomas are extremely rare, the neuroectodermal component undergoing malignant change being a highly exceptional event. To the best of our knowledge, only 18 cases including this one have been described in the literature [1, 2, 4, 6, 7, 8, 9, 10]. Among these cases, 13 were developed within a mature cystic teratoma and 2 within an immature one. Four cases were fibrillary astrocytomas of grade I–II, and 13 were glioblastoma multiform (astrocytoma grade IV). Astrocytic and ependymal tissues, nerve ganglia of sympathetic type, and nerve bundles accompanied by Schwann cells may be seen within mature teratomas as well as cavities lined by choroid plexus, containing cerebrospinal fluid [1]. Whether the neuroectodermal areas of teratomas or glial nodules also contain oligodendrocytes is not known. Several investigators have reported that they were able to detect oligodendrocytes in teratomas, but did not detail defined criteria for their identification [5]. Pilocytic astrocytoma of the central nervous system – a grade-I designated tumor by the World Health Organization – must be distinguished from infiltrating fibrillary or diffuse astrocytoma, the latter being characterized by its capability for tissue infiltration and malignant degeneration [3]. However, to the best of our knowledge, pilocytic astrocytoma arising within an ovarian teratoma has not been previously reported.

It is well known from the literature that gliomas (diffuse astrocytomas and glioblastoma multiform) arising within either mature or immature ovarian teratomas have the capability to settle at ectopic sites, i.e. peritoneal cavity (gliomatosis peritonei) and lymph nodes [4]. Such a possibility would seem unlikely for the present case. Taking into account that the natural history of pilocytic astrocytoma per se is excellent and that, in our case, pilocytic astrocytoma was located almost in the center of the cystic teratoma, without any evidence of invasion or disruption of the cystic wall, no other treatment should be followed. Furthermore, postoperative clinical, laboratory, and imaging studies remain negative. Whether the prognosis of this particular tumor is influenced by its origin cannot be derived from the available literature. The patient has been regularly followed-up and, 8 months after the surgery, is healthy.

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