Virchows Arch (2001) 438:198–200
DOI 10.1007/s004280000293

CASE REPORT

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Meningeal nodules in teratoma of the testis

Received: 23 February 2000 / Accepted: 10 June 2000 / Published online: 3 November 2000

Abstract A case of a 52-year-old man with a mature adult teratoma is reported. Beside histologically mature tissues, this teratoma contained large areas of a meningiomatous proliferation in close proximity of a peripheral nerve and glial tissue. These meningiomatous proliferations were mostly seen in the peripheral parts of the teratoma surrounding the rest of teratomatous elements and were immunohistochemically EMA-positive and S-100 protein- and cytokeratin-negative. Identical meningothelial proliferations are well known in the skin and adjacent soft tissues of the scalp, where they have variously been called sequestrated meningoceles, meningeal hamartoma, cutaneous meningiomas, rudimentary meningocele, hamartoma of the scalp with ectopic meningothelial elements, or cutaneous heterotopic meningeal nodules.

Keywords Testis · Teratoma · Meningeal nodules

Introduction

A teratoma is a benign or a malignant tumor composed of germinal layers: endoderm, mesoderm, and ectoderm. Tumors arising in ectodermal components are relatively rare and, in the literature, a few reports have described primitive neuroectodermal tumors, neuroblastoma, gliomas arising in teratomas of the ovary and testis [1, 2, 6, 9, 13]. We are aware of only one report describing a tumor of meningeal origin in the testis [3]. No such case was ever reported in the ovary or extragonadal sites. We describe a man with an interesting meningeal proliferation arising in a teratoma of testis. Histology of our case differed considerably from the single reported case of gonadal meningioma [3].

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Materials and methods

Clinical history

A 52-year-old man presented with a tumor of the right testis. The tumor was 3.5 cm in size, was well circumscribed, and had a white color and elastic to hard consistency. Macroscopically, it was mostly solid in appearance, with minor areas revealing microcystic change. Five years after the excision, the patient is well, without metastases and without recurrences.

Laboratory methods

The tumor was fixed in 4% formol, routinely processed, and stained with PAS, mucicarmine, and hematoxylin and eosin. Immunohistochemical studies were performed on formalin-fixed, paraffin-embedded sections using the following antibodies: EMA (monoclonal, DAKO), cytokeratin (AE1-AE3, Boehringer), S-100 protein (polyclonal, DAKO), desmin (D-33, DAKO), smooth muscle actin (1A4, DAKO), cytokeratin (CAM 5.2, Becton-Dickinson), glial fibrillary acidic protein (GFAP) (polyclonal, DAKO), synaptophysin (polyclonal, DAKO), and chromogranin (monoclonal, DAKO).

Results

Histologically, the tumor consisted of several types of well-differentiated fetal-type tissues. The well-differentiated teratoma contained areas of smooth muscle proliferation, several types of glandular differentiation, including prostatic, gastrointestinal, and bronchial-type epithelium and cartilage (Fig. 1). On the periphery, there were thick bundles of peripheral nerves intermingled with heavy deposits of syncytial epithelioid meningeal clusters (EMC) (Fig. 2) set in a densely fibrous stroma (Fig. 3). These EMC were PAS- and mucicarmine-negative, and they contained numerous psammoma bodies. I found no mitoses in the EMC. The juxtaposition of the twigs of peripheral nerves and EMC was often very intimate (Fig. 4). By the formation of artifactual clefts around the EMC, these EMC often reminded of foci of metastatic epithelial tumor inside lymphatic vessels (Fig. 5). Small foci of glial tissue were seen in the vicinity of the peripheral nerves and EMC. No immature epi-

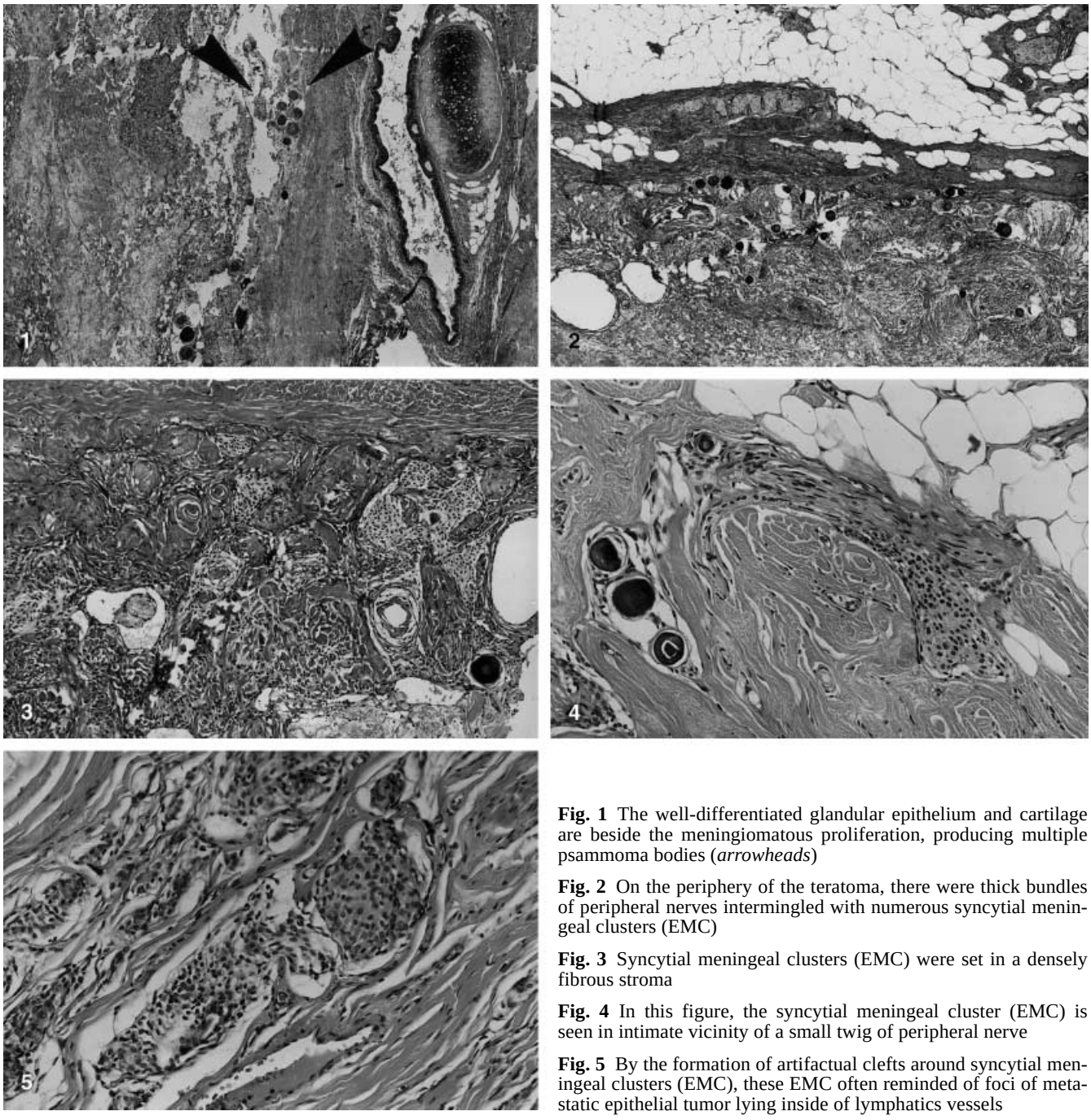

Fig. 1 The well-differentiated glandular epithelium and cartilage are beside the meningiomatous proliferation, producing multiple psammoma bodies (*arrowheads*)

Fig. 2 On the periphery of the teratoma, there were thick bundles of peripheral nerves intermingled with numerous syncytial meningeal clusters (EMC)

Fig. 3 Syncytial meningeal clusters (EMC) were set in a densely fibrous stroma

Fig. 4 In this figure, the syncytial meningeal cluster (EMC) is seen in intimate vicinity of a small twig of peripheral nerve

Fig. 5 By the formation of artifactual clefts around syncytial meningeal clusters (EMC), these EMC often reminded of foci of metastatic epithelial tumor lying inside of lymphatics vessels

thelial, mesodermal, neuroectodermal, or germ-cell tissues were present in the tumor.

Immunohistochemically, the EMC were EMA-positive and were negative with the rest of tested antibodies. S-100 protein stained only the peripheral nerves and glial tissue. GFAP stained only the glial tissue.

Discussion

Teratomas in their pure form account for 2–7.5% of all nonseminomatous testicular neoplasms [7] and represent 20% of mediastinal neoplasms. Tumors arising in the ectodermal component of the teratomas are relatively rare, and only a few reports exist in the literature describing primitive neuroectodermal tumors, neuroblastoma, and gliomas arising in teratomas of the ovary and testis [1, 2, 6, 9, 13]. We are aware of only one report describing a tumor of meningeal origin in the testis [3], and no such case has been ever reported in ovary or extragonadal sites. Our case, however, differed from the case of microcystic meningioma described by Allen et al [3]. These authors described a case of mixed germ-cell tumor, predominantly composed of a mature teratoma with focal

areas of primitive neuroepithelial elements and small foci of seminoma and embryonal carcinoma. The tumor contained a meningiomatous part, which accounted for approximately 15% of the tumor mass. Histologically, this component had the appearance of an autonomous tumor overrunning teratoma, consisting of a microcystic neoplasm devoid of psammoma bodies. The features of the tumor were consistent with a diagnosis of microcystic meningioma seen rarely in intracranial localization [10]. In contrast to Allen et al.'s case, the meningiomatous tissue in our tumor did not form a continuous tumor mass, but consisted of minute epithelioid meningeal clusters (EMC) set in a dense fibrous stroma. At first sight, it reminded more of minute areas of metastatic epithelial tumor than of a neoplastic proliferation surrounding whole tumor in intimate vicinity of thick twigs of peripheral nerve tissue. Identical meningothelial proliferations are well known in the skin and adjacent soft tissues of the scalp. They have been amply published in these locations in the literature, and they have variously been called sequestrated meningoceles [4], meningeal hamartoma [5], cutaneous meningiomas [8], rudimentary meningocele [14], hamartoma of the scalp with ectopic meningothelial elements [15], or cutaneous heterotopic meningeal nodules [16]. Our case thus seems to be the first published case having such meningeal nodules located around a mature teratoma. Our case can be easily differentiated from the foci of squamous-cell carcinomas of metastatic origin or squamous-cell carcinoma arising from teratoma, as rarely occurs in the ovarian teratomas [11, 12], by the lack of proliferative activity, anaplasia, absence of other carcinoma elsewhere in the body, and by the presence of multiple psammoma bodies. Furthermore, our case was EMA-positive and S-100 protein- and cytokeratin-negative, which is an immunophenotype consistent with meningothelial origin and different from carcinoma metastasis. We further believe that the close proximity of the peripheral-nerve tissue and glial tissue around the meningothelial nodules in our case was not fortuitous, but seems to reflect the anatomical close proximity of cranial nerves to arachnoid cells and cerebral glial tissue. In contrast, somatic carcinoma arising in teratoma would probably lack such an arrangement.

Although all tissues in our case were histologically mature, pure mature teratomas are considered to have a potential for metastasis, especially to the retroperitoneal lymph nodes, and because of this fact these patients require continuous surveillance [7].

Acknowledgement I am grateful to Dr. P. Mukensnabl for preparing the photomicrographs.

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