Recurrent chemical meningitis due to an intraspinal cystic teratoma

Case report

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This is the first case report of an intraspinal cystic teratoma that manifested itself in recurrent episodes of chemical meningitis, with no sign of a space-occupying lesion.

KEY WORDS • cystic teratoma • chemical meningitis • congenital tumor • spinal cord tumor

RECURRENT chemical meningitis very rarely constitutes the sole manifestation of an intraspinal tumor; in such case, a dermoid or epidermoid cyst is usually implicated as the cause. Thus far, such meningitis has never been described in conjunction with an intraspinal teratoma. We present a child with such a tumor located at the junction of the conus medullaris and filum terminale. Although he had five distinct episodes of aseptic meningitis, at no time did he present signs or symptoms referable to a space-occupying lesion.

Case Report

This boy, aged 4 years 10 months, was hospitalized for the fifth time with a history of recurrent meningitis. Previously, he had been admitted to another hospital at ages 6 months, 19 months, and 3½ years. He was referred to Hôpital Sainte-Justine when he was 3 years and 10 months old. On each occasion, the symptoms were identical: an abrupt rise in temperature to 39° to 40°C, associated with an intense diffuse headache, vomiting, irritability, and drowsiness. Physical findings were limited to signs of meningeal irritation. There were no signs of any infectious processes. Repeated neurological examination failed to disclose sensory, motor, or sphincter disturbances. The child had never complained of spinal pain.

No abnormality could be detected on spinal examination. On each occasion, temperature and meningismus would subside within 3 to 7 days after onset; between episodes, the child was completely asymptomatic. Past history was otherwise negative.

The results of cerebrospinal fluid (CSF) analysis carried out on each hospitalization are shown in Table 1. Although cultures of the CSF were always negative, appropriate antibiotics were administered intravenously during the first three episodes. On the fourth occasion, however, antibiotics were given only during the first 48 hours.

<table>
<thead>
<tr>
<th>Findings</th>
<th>4/29/74</th>
<th>3/24/75</th>
<th>1/26/77</th>
<th>8/26/77</th>
<th>2/4/78</th>
</tr>
</thead>
<tbody>
<tr>
<td>cells/cu mm</td>
<td>565</td>
<td>3850</td>
<td>875</td>
<td>402</td>
<td>7339</td>
</tr>
<tr>
<td>% polymorphonuclear cells</td>
<td>91</td>
<td>87</td>
<td>94</td>
<td>90</td>
<td>100</td>
</tr>
<tr>
<td>protein (mg/dl)</td>
<td>137</td>
<td>85</td>
<td>30</td>
<td>672</td>
<td>111</td>
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<tr>
<td>(normal: 15–55 mg/dl)</td>
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<tr>
<td>glucose (mg/dl)</td>
<td>38</td>
<td>35</td>
<td>23</td>
<td>69</td>
<td>76</td>
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<tr>
<td>(normal: 70–90 mg/dl)</td>
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Examination. An extensive investigation was carried out when the child was first admitted at Hôpital Sainte-Justine. Skull and spine films, fractionated pneumoencephalogram, computerized tomography (CT) of the brain, tomography of the mastoids, and isotope cisternography failed to disclose any abnormality. On second admission, after a repeated isotope cisternography and CT scan were again negative, a myelogram was performed; it revealed a space-occupying lesion at the level of the conus medullaris. The spinal cord was not displaced, but appeared increased in length and volume; its lower limit was at the level of L-2, and it had displaced the cauda equina roots. There was no evidence of a neurogenic bladder on either the pyelogram or the cystogram.

Operation. A laminectomy was carried out at L1–3. A well encapsulated cystic mass, 1 × 1 × 0.5 cm in area, was situated at the junction of the conus medullaris and filum terminale and attached firmly to the posterior arachnoid. The mass was totally removed. It contained a yellowish creamy liquid. The filum terminale was sectioned 2 cm below the conus medullaris. The postoperative course was uneventful. The child has been symptom-free for the past 16 months; neurological examination is entirely normal.

Pathological Examination. On gross examination, the specimen had a cystic appearance with a smooth internal surface; it contained some grayish granular material. A white firm nodule, 0.5 × 0.4 × 0.3 cm in size, was attached to this cyst. The whole specimen was embedded in paraffin wax. Sections were stained with hematoxylin and eosin, Alcian-blue-PAS, and trichrome.

Histological sections revealed a small firm nodule made of a dense fibrous tissue that contained blood vessels, a few mature fat cells, two fragments of hyaline cartilage, and small cystic cavities lined by a ciliated pseudostratified epithelium and nerve fascicles (Fig. 1). This fibrous tissue also contained small glandular clusters, some of which were of the serous type with PAS-stained intracytoplasmic apical granules, and others were of the mucinous type with Alcian-blue-stained intracytoplasmic vacuoles. A large cyst was attached to this nodule; its lumen contained desquamated cells and a few fibrin strands with polymorphonuclear cells. It was partially lined by a squamous epithelium and partially by a ciliated pseudostratified columnar epithelium (Fig. 2). The cyst wall was made up of fibrous tissue containing bundles of smooth muscle, elastic tissue, and a few duct-like structures lined by two or more layers of transitional cells and filled by a PAS-stained material. In some areas, the lamina propria was infiltrated by neutrophils, lymphocytes, and plasma cells.

Discussion

Most tumors responsible for recurrent meningitis have been dermoid or epidermoid cysts. In the majority of cases, a dermal sinus tract that allowed bacteria to enter the subarachnoid space was present. In the absence of a sinus tract, the contents of the cyst (usually made up of keratin, cholesterol, and lipoid) are discharged into the arachnoid space, giving rise to a leukocytic reaction. Spontaneous cystic spillage, however, is rare, and it has most often been described as a complication of surgery. Instances of other types of tumors causing aseptic meningitis have also been reported: these include craniopharyngioma, hemangioma, and glioma. These tumors, however, were always within the cranial cavity and were responsible for obvious signs of a space-occupying lesion.

Histologically, our case is typical of a well differentiated cystic teratoma. It differs from dermoid or epidermoid cysts because these are lined by a...
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keratinized squamous epithelium; in addition, hair follicles are seen in dermoid cysts. Intraspinal enterogenous cysts, which are located mainly at the thoracic level and are usually associated with vertebral anomalies, have a thin wall; although they are sometimes lined by a squamous and ciliated pseudostratified epithelium, they do not reveal a pericystic firm nodule or cartilage. 6

Teratoma has been defined by Willis 14 as a "true tumor or neoplasm composed of multiple tissues of kinds foreign to the part in which it arises." Many theories 1 have been given concerning its origin; most authors believe teratomas derive from the endoderm of the yolk sac and primordial germ cells. Russell and Rubinstein 11 have stated that teratomas represent the "outcome of perverted development in embryogenesis." If sacrococcygeal teratomas, which constitute a distinct entity, are excluded, cystic teratomas are among the rarest nongliomatous intraspinal neoplasms. 10 In 1978, Takeuchi, et al., 12 extensively reviewed the literature and were able to collect 57 cases: in no instance was meningitis mentioned as a clinical feature. The great majority of the tumors were found in children under 4 years of age. The lumbar or lumbosacral regions were most frequently affected, followed by the cervical region. The tumor could be extra- or intradural, extra- or intramedullary. When situated in the lumbar region (as in our case), the tumor usually adhered firmly to the conus medullaris or cauda equina. It was found to be associated in 20% of the cases with spina bifida and less frequently with other anomalies, such as dermal sinus, syringomyelia, and deformities of the spine. The majority of cases were diagnosed on the basis of the usual complaint of a space-occupying lesion within the spinal canal, motor disturbances and pain being the most frequent symptoms. When radical surgical removal was done, long-term prognosis seemed excellent.

This case report emphasizes the need to perform a myelogram whenever no etiology can be found for recurrent episodes of aseptic meningitis.

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References


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