Postoperative spinal seeding of craniopharyngioma

Case report

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The authors present a case of postoperative spinal seeding of papillary craniopharyngioma. This 27-year-old man who had previously undergone subtotal removal of a suprasellar craniopharyngioma was admitted because of low-back and right leg pain. Results of neurological examination showed a limitation in straight-leg raising in the right side with no sensorimotor changes. Magnetic resonance imaging of the lumbar spine demonstrated multiple enhanced intradural extramedullary masses causing spinal cord compression. Pathological examination of the tumor tissue obtained via laminectomy revealed papillary craniopharyngioma, which had the same histological features as those of the previous suprasellar tumor. Several ectopic recurrences of craniopharyngioma have been reported; however, the authors believe that this is the first published report of the spinal seeding of craniopharyngioma.

KEY WORDS • craniopharyngioma • tumor seeding
moid cysts by the absence of diffuse surface maturation (lamellated keratinous squamae) and the absence of keratothyline granule formation (Fig. 2 upper).

First Postoperative Course. Postoperatively, the patient’s visual acuity had improved; however, postoperative complications such as left-sided homonymous hemianopsia, panhypopituitarism, and diabetes insipidus developed. Subsequently, he underwent gamma knife radiosurgery (20 Gy to the 50% isodose line) for the residual mass. After discharge, his panhypopituitarism, and diabetes insipidus were well controlled with hormone replacement therapy. His visual field defect remained unchanged. In the 14-month follow-up MR image of the brain, there was no evidence of tumor recurrence.

Second Admission

Examination. The patient was readmitted 19 months postoperatively for evaluation of a 2-month history of low-back pain and right radiating leg pain. Results of his admission neurological examination showed a limitation in straight-leg raising on the right side with no sensorimotor changes. An MR image of the lumbosacral spine revealed multiple heterogeneously enhanced masses with cystic portions (Fig. 3 upper and lower left). There was no evidence of a mass on the MR images of the cervical and thoracic spine.

Second Operation. An L1–2 laminectomy and a right L4–5 hemilaminectomy were performed for decompression and histological diagnosis. In the operative field, a gray–pink mass containing yellow cystic fluid was found in the intradural extramedullary portion of L1–2, and a small mass was found among the nerve roots at the level of L4–5. Both lesions showed strong adhesion to the spinal neuraxis and were difficult to remove totally. Subtotal removal was achieved and the patient’s symptoms resolved after surgery.

Histopathological Findings and Second Postoperative Course. The histopathological features of the mass were the same as those of the papillary craniopharyngioma that had been removed previously. It was noted that goblet cells were present in the spinal tumor in addition to the papillary sheets of well-differentiated squamous epithelial cells. There was no evidence of histological malignancy such as nuclear atypism, high mitotic activity, or necrosis (Fig. 2 lower). Subsequent radiation therapy was adminis-
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tered from T-12 to S-3 and the total dose delivered to the lesion for 4 weeks was 45 Gy. The patient was in good condition and free of his symptoms 1 year after spinal surgery and radiation therapy. Follow-up MR images of the lumbosacral spine obtained at that time showed no evidence of tumor recurrence (Fig. 3 right).

Discussion

Total removal of craniopharyngioma is not always possible because of its adherence to important surrounding structures and the resulting risk of considerable morbidity and mortality. The pattern of adherence varies from simple approximation to dense adhesion.\(^1\)\(^2\) Papillary projection of the tumor and reactive gliosis form tight attachments to the hypothalamus, and chronic fluid leakage from the cyst causes adhesion to major arteries.\(^7\) Therefore, craniopharyngioma has a high rate of recurrence even after total removal because of hidden adhesions and/or attachments, and, as can be expected, such recurrence is usually in the vicinity of the primary site.\(^1\)\(^2\)\(^4\)\(^12\)

Seven cases of ectopic and/or remote recurrences have been reported in the literature; these are summarized in Table 1.\(^1\)\(^5\)\(^6\)\(^8\)\(^10\)\(^12\)\(^14\) Among these seven recurrences, five were found along the surgical route and the remaining two both occurred contralateral to the previous craniotomy site and the posterior fossa. The interval between the initial operation and ectopic seeding was between 2 and 25 years; the median was 7 years. Two of the seven cases were adamantinomatous on histological examination, and information on the remaining five was unavailable.

On this basis, one could hypothesize about the nature of a possible mechanism of seeding along the surgical route, because neoplastic cells that contaminate the surgical field during the operation might have the ability to implant and grow.\(^1\)\(^2\) Moreover, the remote recurrence of craniopharyngioma outside the surgical field could be explained by the migration of neoplastic cells spilled into the CSF space due to the dynamics of the CSF. If this is the case, circulation of CSF might also transfer such tumor cells to the spinal canal, from which we infer that the opening of the membrane of Liliequist and the lamina terminalis during surgery might accelerate contamination of the CSF with tumor cells.

Conclusions

We report on a patient with spinal seeding of a craniopharyngioma. We suggest that the spinal seeding of craniopharyngioma through the CSF pathway should be included in the differential diagnosis when a patient who has previously undergone craniotomy for craniopharyngioma develops symptoms of spinal cord compression. Radiation

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Surgical Procedure</th>
<th>Postop Radiation</th>
<th>Site of Recurrence</th>
<th>Time to Seeding (yrs)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Barloon, et al., 1988</td>
<td>needle aspiration via burr hole</td>
<td>done</td>
<td>aspiration tract</td>
<td>2</td>
</tr>
<tr>
<td>Ragoovansii &amp; Pipp graz, 1991</td>
<td>gross-total removal via craniotomy</td>
<td>done</td>
<td>sylvian fissure</td>
<td>2</td>
</tr>
<tr>
<td>Malik, et al., 1992</td>
<td>partial removal via craniotomy</td>
<td>not done</td>
<td>epidural space under craniotomy</td>
<td>20</td>
</tr>
<tr>
<td>Tomita, et al., 1992</td>
<td>partial removal via craniotomy</td>
<td>done</td>
<td>posterior fossa</td>
<td>25</td>
</tr>
<tr>
<td>Israel &amp; Pomezner, 1995</td>
<td>total removal via craniotomy</td>
<td>done</td>
<td>dura under craniotomy</td>
<td>2</td>
</tr>
<tr>
<td>Gupta, et al., 1999</td>
<td>removal via craniotomy</td>
<td>not done</td>
<td>contralateral to craniotomy site</td>
<td>7</td>
</tr>
<tr>
<td>Lee, et al., 1999</td>
<td>total removal via craniotomy</td>
<td>done</td>
<td>sylvian fissure</td>
<td>2</td>
</tr>
</tbody>
</table>
therapy following spinal surgery might be helpful in preventing recurrence of the tumor, as it was in this case. We also maintain that meticulous protection of the operative field and careful handling of the excised tumor during the operation can prevent subsequent spinal seeding.

References
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