Myelopathy due to calcified meningiomas of the thoracic spine: minimum 3-year follow-up after surgical treatment

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Clinical article

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Object. Calcified meningiomas are an uncommon type of meningioma. This study details the clinical features, treatment, and follow-up of 11 calcified meningiomas treated from 2002 to 2009, for the purpose of providing general information, describing the skill required for the surgery, and detailing the imaging study of these tumors.

Methods. Between 2002 and 2009, 11 patients underwent surgery for the treatment of calcified meningiomas. All were treated by the same group of doctors at the same institution, including surgery and rehabilitation after surgery. The minimum 3-year (> 36 months) follow-up data from the 11 patients were detailed. Neurological function was evaluated twice, based on the Frankel scale and Japanese Orthopaedic Association scoring system. The first evaluation occurred before surgery and the second 3 years after surgery.

Results. In 3 cases, the Frankel score decreased by 1 level. In a comparison of the duration of preoperative symptoms, age, degree of canal stenosis, and intraoperative blood loss, it was found that the greater the degree of canal stenosis, the poorer the outcome of the patient. Calcified meningiomas were more likely to adhere to the nerves and dura, a finding that might explain the high incidence of neurological dysfunction and CSF leakage after surgery.

Conclusions. Calcified meningiomas are the most rare of all meningiomas. It appears that a greater degree of canal stenosis can lead to a poorer outcome. Computed tomography scans and MRI with contrast enhancement are recommended for intraspinal tumors before surgery to exclude the possibility of calcification. For calcified meningiomas, precise tumor resection, dura repair during surgery, and medical care after surgery are important for achieving an acceptable outcome.

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Key Words • spinal meningioma • calcification • case series • oncology • Japanese Orthopaedic Association

Spinal meningiomas arise from arachnoid cap cells, which originate in the outer layer of the arachnoid mater and villi. These tumors account for 25%–46% of all primary intraspinal neoplasms.1 Spinal meningiomas can affect people of all ages, but they are most common in those between 50 and 70 years of age.1,14-16 The estimated annual incidence of this type of tumor has been reported to be approximately 5 per million people in women and 3 per million people in men.6 Spinal meningiomas can affect the integrity of nerve function, causing pain and motor dysfunction in patients. Surgical excision remains the treatment of choice. Calcified cases are uncommon and account for only 1%–5% of all spinal meningiomas.5,4 This case series illustrates the clinical features, treatment, and follow-up of 11 calcified meningiomas treated from 2002 to 2009 for the purpose of providing general information, describing the skill required for the surgery, and detailing the imaging study of these tumors. To our knowledge, this study is the first case series of calcified meningiomas.

This article contains some figures that are displayed in color online but in black-and-white in the print edition.
Calcified meningiomas after surgical treatment

Methods

Study Population

The study was a retrospective study. Eleven cases of calcified meningiomas (in 10 women and 1 man) were identified histologically. All cases were treated by the same group of doctors in our department, including surgery and rehabilitation after surgery. The patients ranged in age from 41 to 73 years at diagnosis, with an average age of 58.8 years (Table 1). Five patients (45.5%) were older than 60 years of age. The duration of the preoperative symptoms ranged from 1 to 12 months (mean 7.6 months). All of the lesions occurred in the thoracic region, with 4 lesions occurring at the cervicothoracic junction. Only 1 patient presented with localized pain; the other 10 patients presented with only spinal cord compression, and 1 patient suffered from dysphoria. The neurological status of all patients was classified according to the Frankel score, and the degree of canal stenosis (measured using MRI) ranged from 19% to 73% (mean 35.9%; Table 1). The study was approved by our Institutional Review Board.

Neuroimaging

With calcified meningiomas, the obvious nodular and lamellar high-signal areas can sometimes be observed on CT scans, while there are often no such obvious characteristics on MR images (Figs. 1–3). The masses can be enhanced with injections of intravenous contrast material, according to the report of Lee et al. In that report, between 1998 and 2009, 10 of 11 patients who had pathologically confirmed psammomatous meningiomas showed gross calcifications on CT scans and were included in their study. On MR images, the signal intensity of calcified tumor varied on all imaging sequences. All the tumor masses enhanced after injection of intravenous contrast material (Fig. 4).

Operation and Histopathological Analysis

A posterior approach was performed in all cases, utilizing a posterior screw-rod system combined with autologous or artificial bone grafts to reconstruct the stability of the spine (Fig. 5). Histopathological diagnoses were obtained in all cases. All of the pathological diagnosis results were confirmed by a pathology specialist with training in musculoskeletal oncology (Fig. 6).

Follow-Up Evaluation

All patients returned regularly for clinical and radiographic follow-up evaluations over an average of 55.7 months (range 36–114 months). Patients were sent for radiographic examinations, including radiography and CT or MRI studies, of the surgical margins as well as the adjacent vertebrae. Clinical examinations and radiography were performed 3, 6, and 12 months after surgery during the first year, every 6 months for the next 2 years, and then annually for the rest of the patient’s life. Follow-up data were obtained from office visits and telephone interviews. At 36 months after surgery, neurological function was reevaluated based on the JOA scores and Fran-

TABLE 1: Analysis of 11 cases of calcified meningiomas

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs)</th>
<th>Sex</th>
<th>Location</th>
<th>Duration of Preop Symptoms (mos)</th>
<th>Follow-Up Duration (mos)</th>
<th>Degree of Canal Stenosis (%)</th>
<th>Preop Frankel Score</th>
<th>Postop Frankel Score</th>
<th>JOA Score</th>
<th>Improvement (%)</th>
<th>Adverse Events</th>
</tr>
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<tbody>
<tr>
<td>1</td>
<td>58</td>
<td>F</td>
<td>T7–8</td>
<td>46</td>
<td>1200</td>
<td>1200</td>
<td>D</td>
<td>D</td>
<td>58.8</td>
<td>54.5</td>
<td>recurrence†</td>
</tr>
<tr>
<td>2</td>
<td>57</td>
<td>F</td>
<td>T5–7</td>
<td>46</td>
<td>600</td>
<td>600</td>
<td>D</td>
<td>D</td>
<td>54.5</td>
<td>45.0</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>65</td>
<td>F</td>
<td>T4–5</td>
<td>29</td>
<td>400</td>
<td>400</td>
<td>D</td>
<td>D</td>
<td>50.0</td>
<td>36.7</td>
<td>CSF leak</td>
</tr>
<tr>
<td>4</td>
<td>69</td>
<td>F</td>
<td>T2–3</td>
<td>29</td>
<td>500</td>
<td>500</td>
<td>D</td>
<td>D</td>
<td>66.7</td>
<td>36.3</td>
<td>CSF leak</td>
</tr>
<tr>
<td>5</td>
<td>73</td>
<td>F</td>
<td>T2–3</td>
<td>58</td>
<td>600</td>
<td>600</td>
<td>D</td>
<td>D</td>
<td>73.0</td>
<td>44.4</td>
<td>CSF leak</td>
</tr>
<tr>
<td>6</td>
<td>44</td>
<td>F</td>
<td>T9–10</td>
<td>114</td>
<td>700</td>
<td>700</td>
<td>D</td>
<td>D</td>
<td>25.0</td>
<td>19.0</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>41</td>
<td>F</td>
<td>T4–5</td>
<td>37</td>
<td>500</td>
<td>500</td>
<td>D</td>
<td>D</td>
<td>45.0</td>
<td>28.0</td>
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<tr>
<td>8</td>
<td>39</td>
<td>F</td>
<td>T9–10</td>
<td>32</td>
<td>200</td>
<td>200</td>
<td>D</td>
<td>D</td>
<td>50.0</td>
<td>36.7</td>
<td>CSF leak</td>
</tr>
<tr>
<td>9</td>
<td>56</td>
<td>F</td>
<td>T1–2</td>
<td>29</td>
<td>800</td>
<td>800</td>
<td>D</td>
<td>D</td>
<td>66.7</td>
<td>36.3</td>
<td>CSF leak</td>
</tr>
<tr>
<td>10</td>
<td>56</td>
<td>M</td>
<td>T4–5</td>
<td>58</td>
<td>500</td>
<td>500</td>
<td>D</td>
<td>D</td>
<td>45.0</td>
<td>36.3</td>
<td>CSF leak</td>
</tr>
<tr>
<td>11</td>
<td>63</td>
<td>F</td>
<td>T1–2</td>
<td>28</td>
<td>300</td>
<td>300</td>
<td>D</td>
<td>D</td>
<td>44.4</td>
<td>36.0</td>
<td></td>
</tr>
</tbody>
</table>

* All patients underwent a posterior approach.
† Recurrent meningioma at the first follow-up evaluation.
kel scoring system. A comparison between data from 66 cases with noncalcified meningiomas from 2002 to 2009 and the data in the 11 cases of calcified meningiomas was also provided (Table 2).

Results

All patients had follow-up durations longer than 3 years, and 2 had 5 years of follow-up (Table 1). Intraoperative blood loss ranged from 200 ml to 1200 ml (mean 563.6 ml). No instability of the spine was observed in this case series.

One patient (Case 4) who received a Frankel score of D before surgery suffered from a transient deficit after surgery. Prednisone (1000 mg just after surgery and 500 mg per day) and mannitol (250 ml twice a day) were administered as treatment for neurological dysfunction. The patient began to recover in 2 days, and her neurological function was reevaluated as Frankel score D at her 3-year follow-up.

Four patients suffered from significant incision drainage due to CSF leakage after surgery. After continuous lumbar cistern drainage and frequent dressing changes for 1 or 2 weeks, CSF leakage was relieved. No infection, paralysis, or other serious adverse events occurred in this case series.

None of the 11 cases experienced local recurrence after surgery. One case had recurrent meningioma at the first follow-up evaluation. At the 1-year follow-up evaluation, pain had almost completely disappeared in all of the cases. Spinal cord compression was relieved in most of the cases, but the Frankel scores had not decreased in 8

Fig. 1. Case 5. Images obtained in a 73-year-old woman with a T4–5 calcified meningioma. A: Sagittal MR image showing a mass with low signal intensity. B: Sagittal MR image showing the same mass with enhancement after injection of intravenous contrast material. C and D: Sagittal (C) and coronal (D) 2D reconstructed CT scans demonstrating that the mass was calcified only at the base.

Fig. 2. Case 8. Images obtained in a 65-year-old woman, in which all the tumor was calcified; therefore, complete resection was required. The sagittal 2D reconstructed CT scan (A), sagittal MR image (B), and axial MR image (C) all show a heterogeneous mass.

Fig. 3. Case 10. Sagittal (right) and axial (left) MR images obtained in a 56-year-old man. This meningioma was calcified only at the base, was adherent to the dura, and was resected piecemeal; the calcified part was resected last.
Calcified meningiomas after surgical treatment

No worsening of neurological status was observed at the 1-year follow-up in any of the 11 cases in our series. The comparison between data from the noncalcified cases and calcified cases showed that the JOA score and the rate of adhesion were statistically significant (p < 0.05; Table 2).

The mean values for duration of preoperative symptoms, degree of canal stenosis, and intraoperative blood loss were 7.6 months, 35.9%, and 563.6 ml, respectively (Table 3). The correlation analysis between the duration of preoperative symptoms, degree of canal stenosis, intraoperative blood loss, and outcome measure (JOA score and Frankel score improvement) showed that the correlations between the degree of canal stenosis and outcome measure were statistically significant (p < 0.05; Table 3).

Discussion

The incidence of spinal meningiomas is only lower than that of schwannomas among all primary intraspinal neoplasms. The first successful open laminectomy was attributed to Victor Horsley in 1887; resection and
decompression, therefore, are considered the preferred treatments. Solero et al.14 reported that complete resection was attainable in 97% of meningioma cases. The tumor recurrence rate has been reported to range from 1.3% to 6.4%, spanning a time frame of 1–17 years.6

Spinal meningiomas reportedly occur in women with the greatest frequency (80%) in the posterior, posteralateral, or lateral thoracic region, followed by the anterior cervical region (15%), and most infrequently in the lumbarosacral region (5%).6,13 The incidence of spinal meningiomas in men, however, is 50% in the thoracic region and 40% in the cervical region.17 In this study, all 11 cases of calcified meningiomas occurred in the thoracic region, which increased the risk and difficulty of the surgeries. Calcified cases are uncommon and account for only 1%–5% of all spinal meningiomas.2–4 It is believed that calcified meningiomas are the forerunners of ossified meningiomas. Kitagawa et al.9 concluded that the ossification of meningiomas occurs secondary to metaplasia of arachnoid cells, rather than psammomatous features. Levy et al.,11 in their report of 97 spinal meningiomas, found only 4 cases of calcification. Ossified meningiomas are extremely rare, and a review of the literature revealed only 16 patients with ossified meningiomas from 1932 to 1999, according to the report of Naderi and colleagues.12 Roux et al.13 reported on 3 ossified meningiomas among 54 spinal meningiomas, and they emphasized the functional results of the patients. They reported complete resections in 2 cases and a subtotal resection in 1 case, which required a second surgery and radiation therapy. Both poor and favorable outcomes were reported.

In our study, none of the 11 patients required a second surgery or suffered from recurrence after surgery. Because the follow-up durations in some cases has not been long enough, the possibility of further recurrence cannot be ruled out. Moreover, due to modern surgical techniques, complete resection of tumors is easier than it used to be, and surgery can reduce the rate of recurrence. Nevertheless, 1 patient from this study has suffered a poor postoperative outcome; she must rest in bed and cannot walk on her own. Similarly, in the report of Levy et al.,11 3 of the 4 patients had a “disastrous surgical outcome.”

Sixty-six cases of noncalcified meningiomas were identified in this study, but there were too many confounding factors between noncalcified and calcified cases to conduct direct comparisons, such as the preoperative preparation or the different surgical treatments. The data of these cases are only used to enhance some findings in this study. For calcified meningiomas, the high signal areas on CT are the most important radiological features.

The correlation between outcome measure (JOA score and Frankel score improvement) and other variables, including mean duration of preoperative symptoms, degree of canal stenosis, and intraoperative blood loss, was also described in this study. This analysis showed that there is a negative correlation between outcome measure and the degree of canal stenosis. This finding also suggested that the higher the degree of canal stenosis, the worse the outcome of the patient.

The results of the comparison between the preoperative Frankel and JOA scores and the postoperative scores also show that the improvement in nerve function is not always apparent. This finding could be the result of serious adhesion and spinal cord compression directly caused by the tumor.

When the meningioma is calcified only at the base, especially in cases of adhesion to the dura, the tumor can be resected in a piecemeal fashion, and the calcified part can be resected last (Figs. 3 and 7). When all of a calcified meningioma or the edge is calcified, it is difficult for surgeons to remove it piecemeal, and complete resection is required (Figs. 2 and 7). To remove the tumor entirely an expanded laminectomy procedure should be performed, consisting of bilateral vertebral lamina resection, which we will expand at cranial levels for approximately a half segment, or at caudal levels, or both. Although the operation is performed under a microscope, the effects on the

<table>
<thead>
<tr>
<th>Variable</th>
<th>Noncalcified</th>
<th>Calcified</th>
<th>p Value</th>
<th>Noncalcified</th>
<th>Calcified</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>mean age (yrs) ± SD</td>
<td>53.8 ± 13.08</td>
<td>58.8 ± 9.78</td>
<td>0.227</td>
<td></td>
<td></td>
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<tr>
<td>mean degree of canal stenosis (%) ± SD</td>
<td>31.5 ± 11.81</td>
<td>35.9 ± 16.95</td>
<td>0.284</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>mean JOA score improvement (%) ± SD</td>
<td>64.8 ± 16.14</td>
<td>51.6 ± 14.65</td>
<td>0.013*</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>mean adhesion % (no. of patients)</td>
<td>51.5 (34/66)</td>
<td>90.9 (10/11)</td>
<td>0.015*</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

* Statistically significant.

<table>
<thead>
<tr>
<th>Variable</th>
<th>Correlation</th>
<th>p Value</th>
<th>Correlation</th>
<th>p Value</th>
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<td>duration of preop symptoms (mos)</td>
<td>7.6 ± 4.00</td>
<td>−0.287</td>
<td>0.392</td>
<td>−0.067</td>
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<tr>
<td>degree of canal stenosis (%)</td>
<td>35.9 ± 16.95</td>
<td>−0.619</td>
<td>0.042*</td>
<td>−0.645</td>
</tr>
<tr>
<td>intraoperative blood loss (ml)</td>
<td>563.6 ± 300.91</td>
<td>0.286</td>
<td>0.426</td>
<td>0.260</td>
</tr>
</tbody>
</table>

* Statistically significant.
Calcified meningiomas after surgical treatment

Fig. 7. Photographs of removed calcified meningiomas. Left: Tumor removed in Case 10, a 56-year-old man. The meningioma was calcified only at the base, adhered to the dura, and was resected in a piecemeal fashion, with the calcified part resected last. Right: Tumor removed in Case 8, a 65-year-old woman. The entire tumor was calcified; thus complete resection was required.

nerves, which can lead to neurological dysfunction after surgery, can be avoided in some situations.

Calcified meningiomas are more likely to adhere to nerves and surrounding tissue, especially the dura (Table 2). This complication can be associated with the calcium deposition process of calcified meningiomas. Due to the calcium deposition process, the duration of preoperative symptoms in some cases can appear long. This fact might explain why the dura is sometimes also found calcified. The calcified dura is also removed with the tumor during surgery. In some cases, lateral dura defects, caused by resection of ventral tumors, are more difficult to repair. Tumors should be separated from adhering nerves and from the dura during the operation. These adhesions might be the cause of the high incidence of CSF leakage after surgery.

The tumor should be resected more cautiously because of its adhesions. If dural defects occur because of tumor adhesion, the dura should be tightly sutured. During suturing, the dura should be prevented from being excessively pulled to avoid laceration. The surrounding tissue, such as muscular fasciae, can be used to cover these defects. The dorsal and lateral defects can be repaired, but the ventral defects are too difficult for us to repair. After the operation, intensive medical care and more frequent dressing changes are required. For the patient who suffers from CSF leakage, continuous lumbar cistern drainage of approximately 200 ml per day is recommended. This is believed to reduce the possibility of infection and keep the wound dry so as to promote incision healing. Continuous lumbar cistern drainage is suggested for 1 or 2 weeks. In combination with the frequent dressing changes and medical care for 1 or 2 weeks, CSF leakage was relieved in all patients. For patients suffering from delayed recovery or neurological dysfunction after surgery, rehabilitation will be more important.

In some cases, the spinal cord is compressed by the tumor and looks pale during surgery. After the tumor is removed, ischemia-reperfusion injury is more common and can cause delayed recovery or incurable neurological dysfunction after surgery. For calcified meningiomas, the expanded laminectomy, cautious resection, selected resection, and tight suture are most important during the surgery. Because of the surgical difficulty and poor outcomes with calcified meningiomas, forming a definite diagnosis before surgery is very important. For intraspinal tumors, Lee et al. recommend CT scans and enhanced MRI before surgery to exclude the possibility of calcification.

Mean duration of preoperative symptoms, age, degree of canal stenosis, and blood loss were also compared between the 3 cases in which Frankel scores decreased and the 8 cases in which Frankel scores did not decrease. This comparison showed that no significant difference was detected with regard to age, duration of preoperative symptoms, or blood loss. Furthermore, a similar result was also found after the correlation analysis between the duration of preoperative symptoms, age, degree of canal stenosis, intraoperative blood loss, and JOA score improvement. This analysis suggested that the higher the degree of canal stenosis, the worse the outcome of the patient.

Conclusions

Calcified meningiomas are the most rare of all meningiomas. It appears from this study that a greater degree of canal stenosis can lead to a poorer outcome. Computed tomography scans and enhanced MRI are recommended for intraspinal tumors before surgery to exclude the possibility of calcification. For calcified meningiomas, precise tumor resection, dura repair during surgery, and postoperative medical care are important to achieving an acceptable outcome.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Xiao. Acquisition of data: Xiao, Qian, Wang, Zhang. Analysis and interpretation of data: Xiao, Zhu. Drafting the article: Zhu, Qian. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Xiao. Statistical analysis: Zhu, Qian, Wu. Administrative/technical/material support: Xiao. Study supervision: Xiao.

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