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Two Case Reports of Calcified Spinal Meningioma and a Literature Review

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Study design: Case Report.
Objectives: The aim of this study was to report 2 cases of calcified spinal meningioma that displayed differences in appearance during resection, and to review the current literature on calcified and ossified spinal meningiomas.
Summary of Literature Review: Calcified and ossified spinal meningiomas are rare, and tumor calcification is a risk factor for poor neurological outcomes resulting from the additional manipulations required to dissect the tumor.
Materials and Methods: We describe the clinical course and intraoperative findings of 2 female patients who presented with symptoms of myelopathy. Magnetic resonance imaging showed calcified spinal meningiomas of the thoracic spine. The type of tumor resection performed was dependent on the solidity and texture of the individual tumors.
Results: Pathologic evaluation revealed psammoma bodies, which suggested calcified meningioma. The patients' neurologic symptoms resolved with no neurologic sequelae.
Conclusions: Although there are a few pathologic differences regarding the main type and pathogenesis of ossified and calcified meningioma, both are thought to have a poor prognosis. For these tumors, adequately accounting for the expected poor prognosis and performing a wide laminectomy in order to ensure an adequate surgical margin are important factors for achieving a favorable outcome.

Key words: Calcified meningioma, Ossified meningioma, Myelopathy

Introduction

Spinal meningioma is a common tumor. It represents 25% of all primary intra spinal tumors.[1, 2] Calcified and ossified spinal meningiomas are uncommon, with a reported incidence of 1-5% among all spinal meningiomas.[2-4] The frequency of occurrence of these tumors varies, depending on the spinal level affected[1, 5] ; however, it tends to decrease from the thoracic to the cervical or lumbar spine.[1] Up to 80% of these tumors occur in women of middle age, in the thoracic region.[4, 5] The purpose of our study was to review reported calcified and ossified spinal meningioma cases and to consider radiologic and pathologic findings in addition to the prognosis of these tumors. We also report a rare case of spinal meningioma with an unusual solidity and texture, which was visible with both plain radiography and computed tomography (CT).

Case Report

Case 1

A 51-year-old woman presented with a 2-year history of progressive paresthesia and weakness in both legs. She also displayed a disturbance in gait, that had begun several weeks prior and had progressed since then. Neurologic examination revealed

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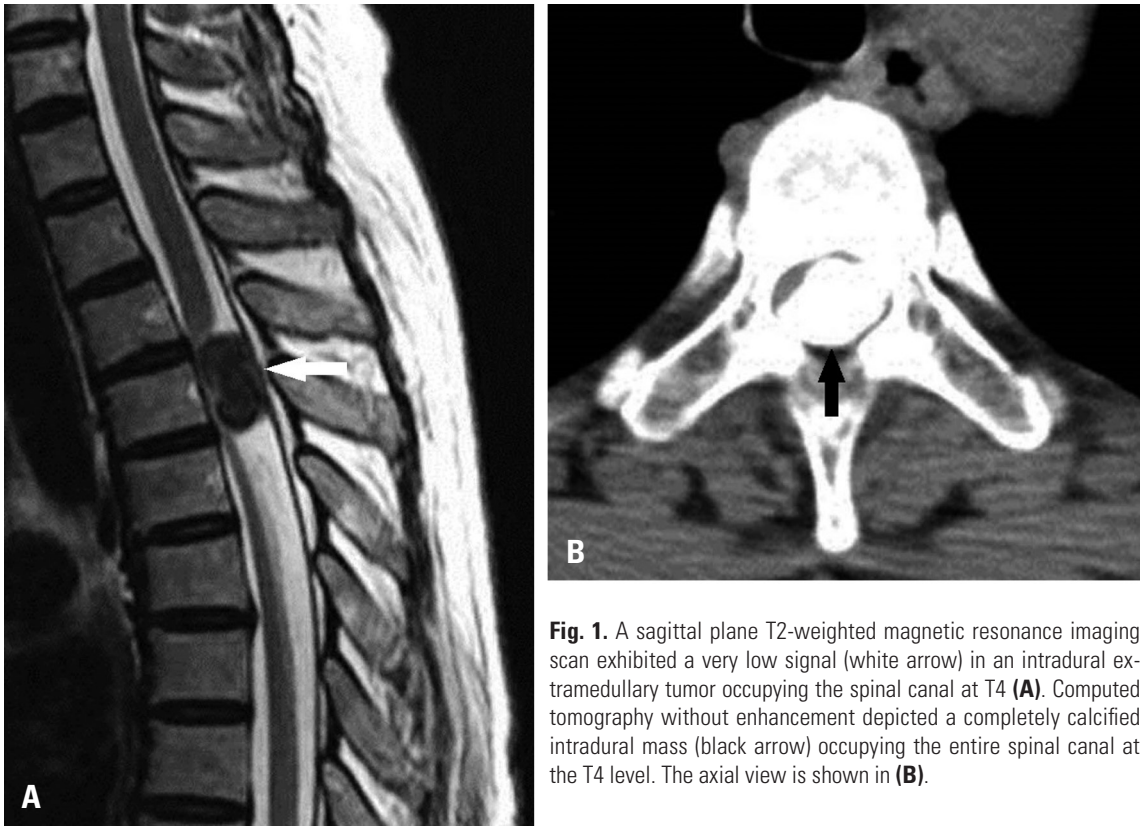

Fig. 1. A sagittal plane T2-weighted magnetic resonance imaging scan exhibited a very low signal (white arrow) in an intradural extramedullary tumor occupying the spinal canal at T4 **(A)**. Computed tomography without enhancement depicted a completely calcified intradural mass (black arrow) occupying the entire spinal canal at the T4 level. The axial view is shown in **(B)**.

decreased muscle strength in the lower extremities, and impaired skin sensitivity, suggesting hypoesthesia below the mammillary line. Deep tendon reflexes (DTR) were increased in both the knee and ankle. A Babinski reflex was obtained, but symptoms of bowel impairment or urinary dysfunction were not observed. Neurologic findings confirmed thoracic myelopathy with mild weakness of the lower extremities, and sensory impairment from below the T4 level. An oblique plain radiograph revealed a large, dense, oval-shaped mass at T4. T2-weighted magnetic resonance imaging (MRI) showed a very low signal in the intradural extramedullary tumor occupying the spinal canal at T4. A CT scan confirmed that the calcified tumor was located in the spinal canal and was severely compressing the spinal cord (Fig. 1).

At the time of surgery, the patient was placed in a prone position and received a general anesthetic. Somatosensory and motor evoked potentials were monitored throughout the entire operation. A midline incision was made in the thoracic region, followed by a wide decompressive laminectomy from T2 to T6. The dura had a relatively firm, hard texture. A midline durotomy was performed using a Potts scissors. The mass severely compressed the spinal cord from posterior to anterior. It consisted of firm calcified tumor tissue, which was adherent to the dura and the arachnoid membrane. However, there was no nerve root involvement and no nerve roots were sacrificed. The tumor and its adhesion to the dura were meticulously dissected. In spite of attempts to

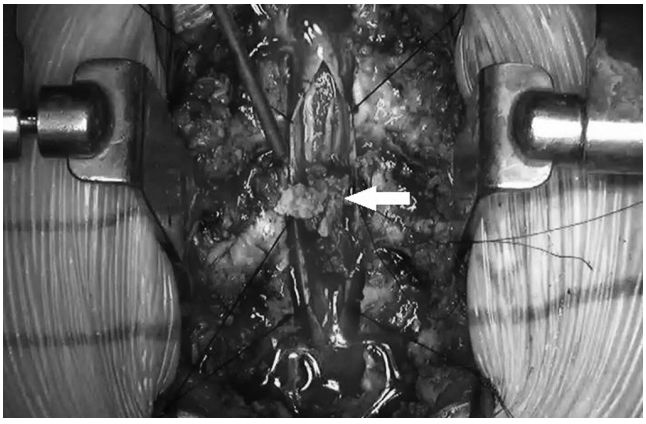

Fig. 2. Intraoperative image showing a tumor (white arrow) at the T4 level. The tumor had a friable solidity and was dissected in a piecemeal manner.

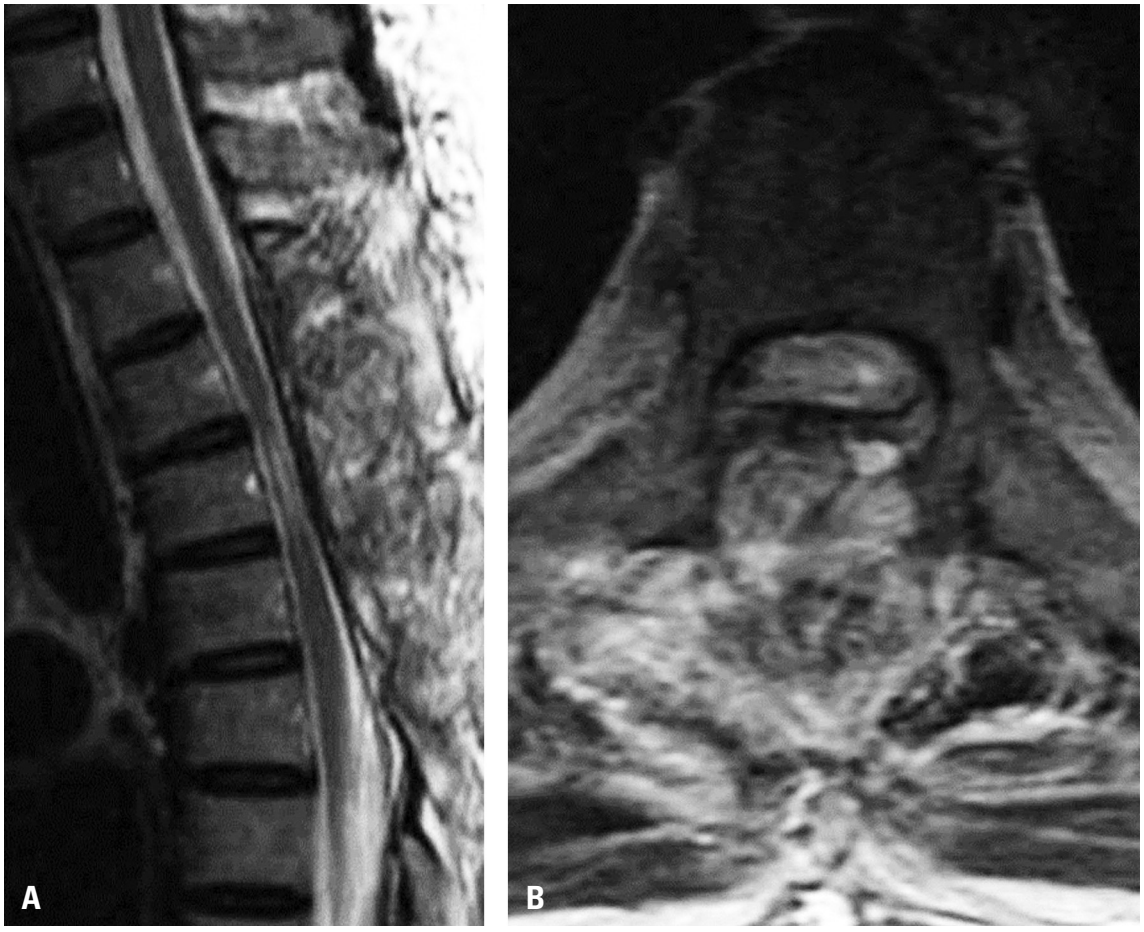

Fig. 3. Postoperative T2-weighted magnetic resonance imaging scan revealing decompression of the flattened cord and recovery of the cord diameter in the thoracic spine. The sagittal view is shown in **(A)**, and the axial view in **(B)**.

perform the mass resection en bloc under the microscope, it was impossible to remove it in this manner due to the soft, friable condition of the mass (Fig. 2). The mass was divided into several pieces, which were removed by piecemeal resections. Despite the severe adhesion of the dura and tumor, there was no defect in the dural membrane, and its repair was performed using a 6-0 Prolene suture. Decompression of the flattened medullary ring was revealed on postoperative T2-weighted MRI of the thoracic spine (Fig. 3). Histopathological examination of the removed specimen by hematoxylin and eosin staining revealed a psammomatous meningioma with abundant psammoma bodies and areas of micro-calcification, suggesting a calcified meningioma (Fig. 4). Neurologic symptoms subsided dramatically and 2 weeks after the surgery, the patient was ambulatory without any disturbance in gait. The patient was discharged from hospital and returned to her daily life.

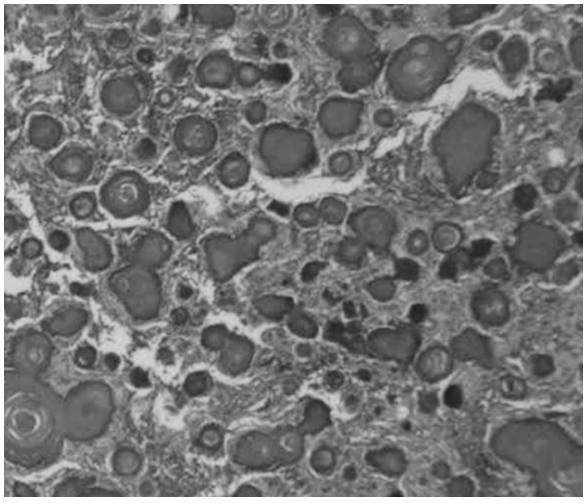

Fig. 4. Microscopy of the specimen demonstrating a psammomatous subtype that was composed of numerous calcified psammoma bodies, suggestive of a calcified meningioma (hematoxylin and eosin staining, ×200 magnification).

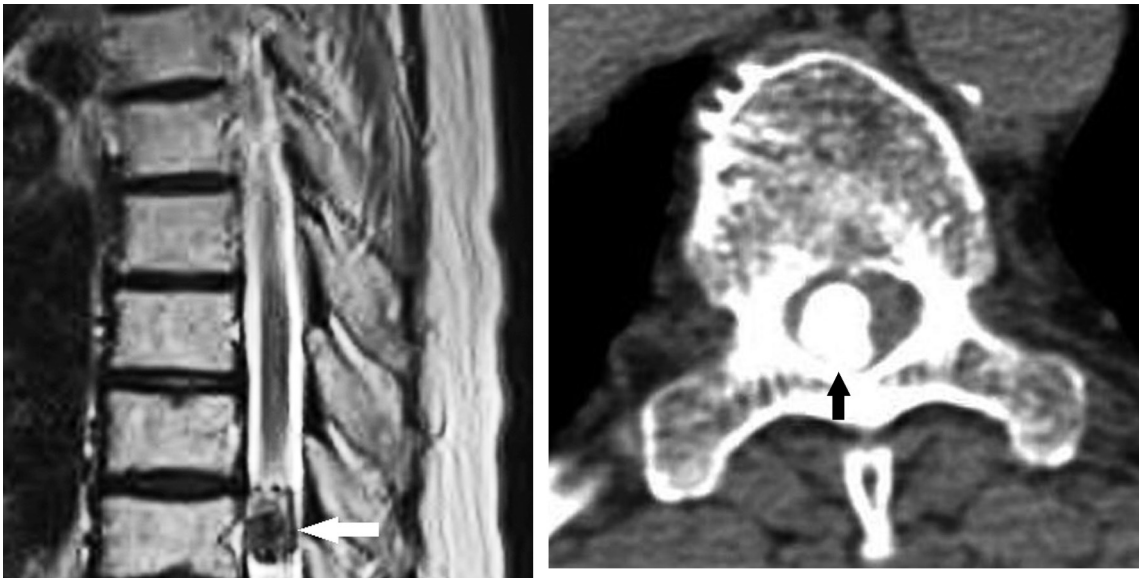

Fig. 5. T2-weighted magnetic resonance imaging scan showing an oval-shaped mass (white arrow) clearly demarcated by a hypodense signal. The sagittal view shown in **(A)**. Computed tomography image showing a calcified mass (black arrow) occupying the spinal canal at the level of the T9 vertebra. The axial view is shown in **(B)**.

Case 2

A 77-year old woman presented with intermittent back pain and neurologic symptoms, which included numbness and a progressive ascending weakness of the lower extremities. Paresthesia and grade 4/5 muscle weakness of the lower extremities were detected on physical examination. DTR was normal and pathologic reflexes were not observed. An oval-shaped calcified mass at the T9 vertebra level was apparent on a plain radiograph. The sagittal plane of a T2-weighted MRI scan showed an area of hypodensity at T9; on axial view, the mass was seen to occupy the spinal canal. Additionally, an oval-shaped calcified mass (2.5 cm in size) could be seen on a sagittal view CT scan (Fig. 5). Anesthesia and surgical position were as reported in the previous case. A wide laminectomy below and above T9 was performed. The dura had a hard, firm texture due to the influence of the mass. Adhesions between the dura and the mass, as in the preceding case, were meticulously dissected. When attempts were made to resect the mass, it exhibited a consistency different to that reported for the previous case; it was not friable and was actually quite firm. Mass resection was performed slowly, and total resection was accomplished en bloc, using an operating microscope at high magnification (Fig. 6). Repair of the dura was accomplished without damage to the dural

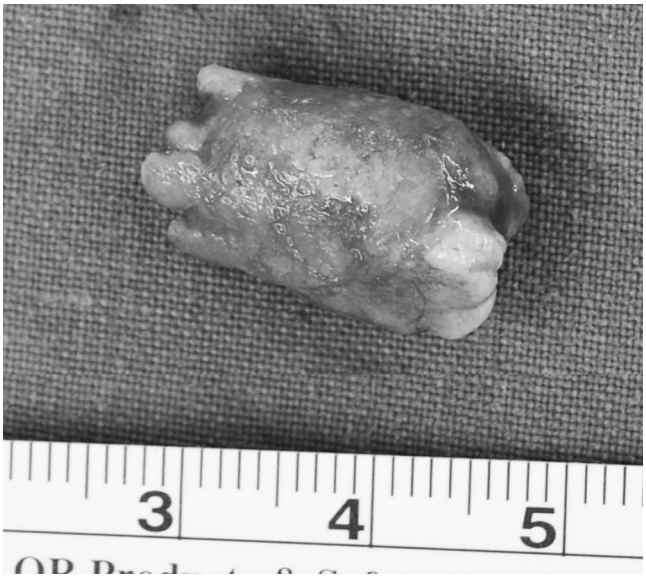

Fig. 6. Gross photography showing en bloc dissection of the firm, solid tumor.

membrane. Two weeks after surgery, a T2-weighted MRI revealed the absence of the hypodense area, and recovery from neural encroachment. Postoperative histologic examination revealed a psammomatous meningioma, as in the previous case.

Discussion

The discovery of calcification in the tumor lesions is crucial for evaluating the potential difficulty of resection and the expected prognosis after the operation in addition to ruling out the differential diagnosis. Calcified, ossified meningioma can be visible with plain radiography, when appears to contain bone or calcified psammoma bodies, resulting in a hard tumor.[2] However, it cannot be identified with plain radiography in all cases; visualization of calcified meningioma of the spinal canal with plain radiography has been reported in just 1-5% of cases. Among the 26 reviewed cases, only 2 cases of calcified meningioma were described as being visible on plain radiography. CT and MRI with contrast enhancement are usually recommended for intraspinal tumors prior to surgery, to exclude the possibility of calcification.[3] Even if MRI is considered the best non-invasive neuroimaging technique for the prevention of misdiagnosis, it cannot detect small amounts of calcification in all cases. Furthermore, calcifications generally display low signal intensity on MRI. For calcified, ossified meningiomas, the high signal areas on CT are the most important radiological features.[4] Although ossification or calcification of spinal meningioma are mainly depicted as very low density areas on T2-weighted MRI and relatively low density areas on T1-weighted MRI, some cases reveal different densities on MRI. On reviewing the cases with MRI findings, we found that they tended to show very low density on T2-weighted images and relatively low density on T1-weighted images. However, the T1-weighted images of these tumors were described as showing mixed intensity or high intensity, according to the authors. In one case, reported by Niijima et al,[6] homogeneous high signal intensity on T1-weighted MRI was demonstrated. We agree with previous reports that the intensity of calcified and ossified regions can be varied and nonspecific, especially on T1-weighted MRI images. In our case, it was possible to identify the mass boundary, and to confirm calcification through CT. Therefore, it enabled us to expect some surgical difficulty prior to embarking on the resection. In the reviewed cases, all calcified or ossified spinal meningioma instances revealed higher density on CT scans, similar to that of bone. CT is considered an essential diagnostic tool in ossified or calcified spinal meningioma for the delineation of the exact location of the lesion and the identification of surgical margins.

Calcified meningioma is believed to be the forerunner of osteogenic meningioma.[7] Common histologic subtypes of ossified meningioma have been identified as transitional, psammomatous, and metaplastic meningiomas Many authors have made suggestions about the possible mechanism that leads to ossification and calcification of spinal meningioma. Uchida et al, suggested that both ossification and calcification were induced through exposure of the area to the biochemical activity of the ossification cascade, or to osteoblast transforming factors like Sox9 and Runx-2, secreted from premature mesenchymal cells.[8] Kubota et al, describe the formation of psammoma bodies. They are initially induced by hydroxyapatite crystals, which gradually aggregate within the bodies. Next, collagen fibers surrounding the calcified bodies start to accumulate deposits of apatite crystals. As a result, huge psammoma bodies are formed.[9] Additionally, Doita et al, report that calcified psammomatous bodies may not always lead to bone formation.[2] Some authors have suggested that ossification of meningiomas occurs secondary to the metaplasia of arachnoid cells, rather than psammomatous features. In the case reported by Huang et al, the described bone histogenesis was different from that of psammomatous cases.[7] Here, only lamellar bone with bone marrow appeared in the transitional meningioma, with no features of reactive bone formation or invasion. According to Chotai et al, 2 out of 9 reported cases, and the present case of ossified spinal meningioma, contained fatty or hematopoietic marrow in the ossified tissue.[10] These findings suggest that, even in the psammomatous subtype of ossified meningioma, the metaplastic differentiation of neoplastic cells to osseous and hematopoietic tissue might play a crucial role.

The prognosis of spinal meningioma is generally favorable. According to Solero et al, approximately 90% of spinal meningiomas can be surgically resected. The rate of recurrence with long-term follow-up is reported as ranging from 4% to 10%.[2] Tumor calcification is a risk factor for poor neurological outcome as a result of the additional manipulations required to dissect the tumor.[5] Furthermore, there is a higher recurrence rate with calcified or ossified spinal meningioma. Of the 26 reviewed cases, 9 had neurological involvement, suggesting that these types of meningioma have a worse prognosis than the other types mentioned previously. In 11 cases of calcified- meningioma and

15 cases of ossified meningioma, neurologic complications were reported in 5 and 4 cases, respectively. According to Schaller et al, resection of spinal psammomatous meningiomas is associated with a less favorable neurological outcome than resection of spinal meningiomas of other subtypes. In a study of 4 cases of calcified meningioma by Levy et al, 3 out of the 4 patients had a poor prognosis. Although Niijima et al. reported that the outcome after total microsurgical removal was good in either calcified or ossified early stage spinal meningioma,[6] many studies have reported cases with neurologic symptoms. Regardless of the technical assistance provided by the microscope, ossified and calcified meningiomas are very difficult to remove in a piecemeal manner, and it is very difficult to separate the mass from the attached dural membrane. In a publication by Zhu et al, the authors outline the factors necessary for achieving an acceptable outcome in cases of calcified meningioma. These factors include precise tumor resection, dura repair during surgery, and medical care.[4] Chotai et al, suggested that subtotal resection, the presence of extradural components, young age, multiple lesions, calcification, ossification, and an anterior location were prognostic factors associated with a relatively poor outcome.[10] Calcified cases demonstrated a poor Japanese Orthopedic Association score and higher adhesion rate compared to non-calcified meningioma.[4] In calcified or ossified meningiomas, there is usually adhesion of the tumor to the surrounding nerve tissue, making resection difficult, and possibly contributing to a worse surgical outcome. The two masses reported here exhibited different solidity and texture, although both cases had the same preoperative and postoperative diagnosis and exhibited similar image findings. Even with a preoperative diagnosis made using MRI and CT, it was not possible to predict the degree of dural adhesion or mass features, such as texture and solidity, that would determine the level of surgical difficulty. Both the choice of resection technique, either piecemeal or en bloc, and the postoperative results can be affected by features of the mass that are identified intraoperatively. In both of our cases, it was advantageous that the total laminectomies were performed as wide as possible to facilitate easier adhesion detachment and mass removal.

Conclusion

In summary, although there are a few pathologic differences regarding the main type and pathogenesis of ossified and calcified meningioma, both are thought to have a poor prognosis.

Hypointensity on T1- and T2-weighted MRI should prompt the surgeon to expect calcified lesions, which may be associated with calcified or ossified meningioma. We believe that CT enables the identification of the exact boundaries and lesions of the mass. For these tumors, an expected poor prognosis and a wide laminectomy that can acquire an adequate surgical margin are important factors in order to achieve a favorable outcome.

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석회화된 척추 뇌수막종 증례보고 및 문헌검토

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연구 계획: 증례 보고

목적: 석회화된 척추 뇌수막종 2개의 증례에 대해 보고하며, 최근 석회화, 골화된 척수 뇌수막종에 대한 문헌을 검토하고자 한다.

선행문헌의 요약: 석회화, 골화된 척추 뇌수막종의 발생빈도는 드물며, 수술적 치료시 제거과정에 과도한 조작으로 인해 일반적인 척추 뇌수막종에 비해 예후가 나쁘다고 알려져 있다.

대상 및 방법: MRI 상 흉추에 석회화된 척수 뇌수막종을 가진 두명의 여성환자에 대한 수술적 제거를 시행하였다. 수술적 제거술시 종양의 견고함과 조직양상은 각각 달랐다. 적절한 수술적 경계를 확보하며 광범위한 추궁판 절제술도 시행하였다.

결과: 병리학적 소견상 석회화된 사립체(psammoma body)을 보였다. 환자의 신경학적 증상은 호전되였다.

결론: 석회화, 골화된 척추 뇌수막종의 수술적 치료는 예후가 나쁘다고 알려져 있다. 이러한 종양제거술시 적절한 수술적 경계를 선택하여 시행한 광범위한 추궁판 절제술은 술후 좋은 예후를 얻을 수 있는 중요한 인자이다.

색인 단어: 석회화된 뇌수막종, 골화된 뇌수막종, 척수증

약칭 제목: 석회화된 척추 뇌수막종 증례보고

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