Completely ossified thoracic intradural meningioma in an elderly patient
A case report and literature review

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Abstract

Rationale: Spinal meningioma is the second most common spinal neoplasm that commonly occurs in middle-aged women. As a rare histological variation of meningioma, completely ossified meningioma (OSM) and its standard surgical strategies have been reported. However, the surgical outcomes of elderly OSM cases (age >70) are less investigated. Herein, we intend to present an elderly OSM case and review the elderly OSM cases in published literatures.

Patient concerns: An 85-year-old female presented with 10-year history of back pain and developed radiating pain on her left lower extremity within the last 2 weeks.

Diagnoses: A magnetic resonance imaging scan revealed an intradural tumor at the T11 level. A computed tomography scan demonstrated that the mass was completely ossified and had distinct borders (Fig. 1D–F). In a dynamic contrast-enhanced magnetic resonance imaging scan, the mass presented as a lateral intradural extramedullary tumor with abundant blood supply (Fig. 2).

Interventions: The patient underwent en bloc intradural tumorectomy via posterior approach.

Outcomes: After surgery, the patient’s pain was relieved. However, the patient spent another 3 weeks in hospital due to a series of post-surgery complications, including hypostatic pneumonia, pulmonary heart failure, hydrothorax in both thoracic cavities, hypoproteinemia, and deep venous thrombosis on both of her legs. The complications recovered after 3-weeks treatment. In 1-year follow up, no additional symptom was found and the patient was recurrence free.

Lessons: Our report indicated that the surgical outcome can be satisfying in elderly OSM patients, while the post-operative complications frequently occur due to the poor physical condition of elderly patients. As a result, treatment of peri-operative complications of elderly OSM patients also deserves greater attention along with surgical resection.

Abbreviations: CF = cerebral fluid, CT = computed tomography, DVT = deep venous thrombosis, MRI = magnetic resonance imaging, OSM = completely ossified meningioma.

Keywords: completely ossified meningioma, elderly patient, en bloc tumorectomy, post-surgical complication, thoracic spine tumor

Editor: Maya Saranathan.
FX, ZT, and ZQ contributed equally to this work.
This study was supported by Education Department of Jilin Province (No. JJKH20190034KJ, Dr Yuanyi Wang).
This study was approved by the Ethics Committee and the Institutional Review Board of the First Hospital of Jilin University.
Patient consent was approved and signed by the patient herself.
Patient has provided informed consent for publication of the case.
The authors have no conflicts of interest to disclose.
The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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How to cite this article: Xu F, Tian Z, Qu Z, Yao L, Zou C, Han W, Zhang C, Fu C, Wang Y. Completely ossified thoracic intradural meningioma in an elderly patient: a case report and literature review. Medicine 2020;99:25(e20814).
Received: 8 December 2019 / Received in final form: 24 April 2020 / Accepted: 21 May 2020
http://dx.doi.org/10.1097/MD.0000000000020814
1. Introduction

Spinal meningioma accounts for 25% to 46% of all cases of primary spine tumors. In terms of frequency, meningioma is the second most common neoplasm that occurs in the spine (neurilemmoma being the most common neoplasm). Calcification and ossification of spinal meningioma are uncommon, with an incidence of 1% to 5% among all cases of spinal meningioma. Most cases of meningioma occur among middle-aged women, whereas completely ossified meningioma (OSM) and calcified meningioma have been reported as rare histological variations of meningioma. OSM has been considered as an advanced form of calcified meningioma, and its diagnosis and treatment have been studied and reviewed in recent years. A standard surgical strategy of tumor resection has been widely accepted by clinicians. However, the surgical outcomes of elderly (age >70 years) cases of OSM are seldom reported. Because the ossification process occurs slowly, elderly patients with OSM also present with clinical symptoms, and subsequent surgery might lead to serious complications because of their age. Here, we present a case of a patient with OSM, who was also the second oldest patient with OSM in the world.

2. Case report

An 85-year-old female presented to the spine surgery clinic with a 10-year history of back pain; she had developed radiating pain on her left lower extremity within the last 2 weeks. During the physical examination, the patient was restricted in a seated position and had back tenderness and percussion. Her muscle force of both lower limbs was decreased (level 3), combined with significant sensory loss below the groin level. Her tendon reflexes were normal, and she had negative Babinski reflexes on both sides.

A magnetic resonance imaging (MRI) scan revealed an intradural tumor at the T11 level that caused serious compression of the thoracic spinal cord (Fig. 1A–C). A computed tomography (CT) scan demonstrated that the mass was completely ossified and had distinct borders (Fig. 1D–F). In a dynamic contrast-enhanced MRI scan, the mass presented as a lateral intradural extramedullary tumor with abundant blood supply (Fig. 2).

After pre-operative preparation, the patient underwent T9 to T12 transpedicular screw fixation, T10 to T11 laminectomy, unilateral facetectomy, and intradural tumor resection via a posterior approach. After the removal of laminar and facet elements of T10 and T11, the dura matter of the corresponding level was sufficiently exposed. A dura incision was made, and the extramedullary mass was revealed, unlike the rocky impression that was presented in the CT scan, and the lesion surface was tough and fibrotic (Fig. 3). The mass entity was closely wrapped by nerve tissues (Fig. 3), and the ventral side of the mass had serious adhesion to the arachnoid membrane and spinal cord. After the tumor base and the attached ventral arachnoid membrane were removed, the spinal canal was sufficiently decompressed. The patient had a smooth recovery, and the sensory and motor function of her lower limbs improved during the follow-up period.
membrane were carefully separating using bipolar forceps, the mass was completely resected (Fig. 3) while the spinal cord was cautiously avoided, and the dura was closed with sutures afterwards. The operating time was 2 hours and 20 minutes, and the total blood loss was approximately 400mL. A histopathological examination of the resected specimen revealed that the mass was a meningioma with diffused psammomatous bodies (Fig. 4).

The post-surgical examination reported complete removal of the calcified tumor and appropriate instrument location (Fig. 5). The radiating pain was relieved immediately after the surgery; however, the patient mentioned slight involuntary movement on her left lower extremity, which improved after three weeks of neurotrophic treatment. She stayed for 3 weeks in the hospital due to the perioperative complications. Chronologically, the patient had cerebral fluid (CF) leakage (300mL/d) due to the absence of biomaterial product that could reinforce the dura suture. During the period of bed stay without pillow, which prevented severe CF leakage, she further developed hypostatic pneumonia, which leaded to pulmonary heart failure and hydrothorax in both thoracic cavities. Moreover, she had hypoproteinemia, and deep venous thrombosis (DVT) in both of her legs because of albumin loss and chronic consumption and long-time bed stay. She received antibiotic therapy (cefodizime, 20mg/BID), sputum excretion, thoracic drainage, and anti-thrombosis therapy and was administered a protein-rich diet and albumin intravenous drip. After 3 weeks, her blood protein level increased, and the hydrothorax and cardio-related symptoms were subsequently relieved. Then, her DVT transformed to chronic thrombus, and she was discharged from the hospital three weeks post-surgery. During the 1-year follow-up, the patient did not have any additional symptoms and was recurrence-free.

3. Discussion

Including the current case, a total of 10 cases of elderly (age > 70 years) patients with OSM have been published up till 2019 according to PubMed (Table 1). The average age was 78 years, and our patient is the second oldest patient who underwent OSM tumorectomy. According to the published case reports, the predominant incidences among females have been clearly noted among elderly patients (9 women and 1 man), and most cases of OSMs (9 cases) occurred in the thoracic spine, with only one exception that occurred at L3. Interestingly, most patients did not report a symptomatic history over 2 years, and several cases remained asymptomatic until an event such as falling occurred. According to the onset peak and age range of the cohort, the mass was speculated to have been asymptomatic or slightly symptomatic for a long time; the mass then became unstable under stimulation, which might result in bleeding, displacement, and fracture of the mass, thereby shifting and causing nerve compression and aggravated nervous symptoms. For treatment, all of the reviewed cases underwent tumor resection, in which the tumor was removed in 6 cases via total en bloc resection, and 3 cases’ tumor was removed with adherent dura matter. All of the patients showed immediate or gradual improvement after surgery, and no recurrence was noted in the follow-ups.
| Citations                        | Age (yr) & gender | Level | History | Neurological deficit | Treatment | Postoperative course                                                                 | Unfavorable events and peri-operative complications | Time to discharge | Follow-up time |
|---------------------------------|-------------------|-------|---------|----------------------|-----------|--------------------------------------------------------------------------------------|------------------------------------------------------|-------------------|----------------|
| Niijima et al, 1993[6]          | 75 F              | T8–9  | 1 yr    | Ataxic gait, painless progressive numbness in lower extremities, unable to walk | Tumorctomy with dura attachment | The patient's neurologic deficits were gradually improved. | Mild paraparesis and residual numbness.                  | 14 mo             |                |
| Nakayama et al, 1996[7]         | 74 F              | T9    |         | Progressive paraparesis and hypesthesia in lower extremities | Surgery   |                                                                                      |                                                       |                   |                |
| Huang et al, 1999[8]            | 73 F              | T5    |         | Back pain, paraparesis, and urinary incontinence | Tumorctomy | Neurologic deficits were improved.                                                   |                                                      |                   |                |
| Liu et al, 2006[9]              | 70 F              | T11   | 1 wk    | Back pain, paraparesis, and urinary incontinence | En bloc tumor resection | Neurologic deficits were resolved.                                                  | Had stroke 2 mo post-surgery.                        | 2 yr              |                |
| Hirabayashi et al, 2009[10]     | 82 F              | L3    | 1 yr    | Progressive pain and numbness in lower extremities, urinary incontinence | En bloc tumor resection | Neurologically intact, recurrence-free in 5 yr.                                      |                                                      | 5 yr              |                |
| Uchida et al, 2009[11]          | 76 F              | T8 and T11-T12 | 2 yr | Painful muscle weakness and numbness in lower extremities | En bloc resection with parts of the dura mater and arachnoid. | Remarkable neurological improvement without complications. |                                                      | 10 d              | 2 yr           |
| Taneoka et al, 2013[12]         | 78 F              | T9    | 3 mo, aggravated in 2 mo | Progressive pain in lower extremities | Tumorctomy with the inner dura | The patient's neurologic deficits were improved. |                                                      |                   |                |
| Xia and Tian, 2016[13]          | 90 M              | T10-11 | 1 wk after a fall Back pain and paraplegia | Tumorctomy | En bloc tumor resection | The patient's neurologic deficits were gradually improved. |                                                      |                   |                |
| Kim et al, 2016[14]             | 77 F              | T9    |         | Back pain, numbness and a progressive ascending weakness | Total en bloc tumor resection | The patient's pain improved, post-surgical complications recovered within 3 wk, and recurrence-free in 1-yr follow-up. | Central fluid leakage, hypostatic pneumonia and deep venous thrombosis | 3 wk              | 1 yr           |
| Present case                    | 85 F              | T11   | 10 yr history, aggravated in 2 wk Back pain and radiation pain on her left lower extremity. | Total en bloc tumor resection |                                                       |                                                      |                                                       |                   |                |

Including the present case, only 9 cases of elderly completely ossified meningioma patients were reported since 1993, among which, 8 cases were documented in English and 1 case was reported in Japanese.

F = female, L = lumbar, M = male, T = thoracic.
Calciifications in cases of spine meningioma are rare, and gross calcification was only observed in 1% to 5% of all cases of spinal meningioma.10 Cases of completely ossified spinal meningioma are even rarer. The classification by the World Health Organization (WHO) states that the histological classification of ossified meningioma has been categorized as a phenotype of meningioma with mesenchymal component expression.11 Based on the calcification degree of meningioma, Ruggeri et al proposed a classification that included 3 subgroups: micro-calci fized, macro-calci fized, and ossified. In Ruggeri’s classification, meningiomas that only have histological evidence of calcification are categorized as micro-calci fized; macro-calci fized meningiomas and ossified meningiomas refer to a mass that has gross calcification in part or in total, respectively, which can be identified in a CT scan.16 This classification clarified the distinction of calcification and ossification of meningioma.

Currently, researchers agree that meningioma is a form of dysplasia that arises from arachnoid cap cells originating from the outer layer of the arachnoid mater and villi;12 however, the pathogenesis of OSM is still controversial. Based on the histological presentation of calcified meningioma, WHO has described three common histological subtypes of OSM, including transitional, psammomatous, and metaplastic.17 In a study by Kubota et al, the progression of psammomatous body formation was described. According to their theory, ossification was presented as an advanced and large calcification of the piamaterous body with the progressive accumulation of a hydroxyapatite crystal,18 which explained the formation of the psammomatous body in our case. Another study proposed that the ossification started from metaplasia of arachnoid cells including premature arachnoidal cells and meningotheial cells, which induce the synergistic effect of osteoblast, fibroblast, and angiogenesis components in bone tissue formation, thus promoting the ossification in meningioma.19 Uchida et al suggested that the calcification and ossification progress is induced by the activation of the ossification cascade or exposure to osteoblast transforming factors including SOX9 and Runx-2.20 However, along with the findings of bone histogenesis and bone tissue in cases of OSM,19 researchers have begun to realize that the metaplastic differentiation is also critical in all subtypes.

The diagnosis of calcification and ossification meningioma is not difficult since the lesion sometimes can be observed in a digital radiograph (DR), and more easily identified in a CT scan, which is also the sorting method of macro-calci fized meningioma and ossified meningioma.16 MRI is helpful when evaluating the anatomical localization and neurovascular involvement of neurogenic tumors including meningioma. Usually, OSM appears as hypo- or isointense signals in the spinal cord on T1- and T2-weighted sequences with hypointense signals in calcified areas. Furthermore, the OSM signal can be enhanced by contrast, which requires a differential diagnose apart from other tumors including schwannoma, lymphoma, and metastasis.20,21 However, MRI is not as sensitive as CT in detecting small amounts of calcification, and the uncertain signal (mixed or high density) of calcification presented in a T1-weighted sequence further increases the difficulty in identifying calcifications via MRL. Therefore, CT has been considered as an important tool in OSM to delineate the location and margin of the mass.7

For benign meningioma, surgery is the primary treatment for OSM, and a satisfying outcome can be expected when the mass is completely removed. However, unlike typical spinal meningiomas, OSM usually adheres to surrounding nerve tissues. The pattern of adhesion might be attributed to its scant cellularity consisting of acellular concretions.21 Especially for elderly patients, the lack of a clear dissection plane due to the metaplasia of surrounding arachnoid membrane might result in surgical difficulties and risks that lead to worse surgical outcome compared to usual meningiomas.22 Thus, the prime goal of surgery is further tumor resection with careful maintenance of nerve tissue.23 In our experience, OSM resection for elderly patients is performed with several principles. First, a thorough and detailed pre-surgical preparation is necessary. The common underlying diseases of the circulation and respiration systems should be properly treated until the patient is able to tolerate general anesthesia. The mass’s location and size should be fully estimated, and the minimal range of laminectomy must be discussed by the surgical group. Secondly, the duration of the surgery must be minimal, which results in less anesthetic risks and blood loss. Thirdly, considering the age and recurrent rate of OSM among elderly patient, if the base of the OSM closely adheres to the surrounding nerve tissue, which requires a rather long time to resect it entirely, the base adhesion is better coagulated or directly debulked with the attached inner dura layer than separating the adhesion, thereby reducing the risks of anesthesia and nerve injury. At last, the dura matter has to be carefully closed to prevent serious cerebral leakage after the surgery.

According to published studies, favorable surgical and satisfying follow-up outcomes have been reported (Table 1); nonetheless, post-surgical complications of elderly patients with OSM have been insufficiently documented. The perioperative complications are seldomly referred among the reported elderly OSM cases, only 1 case was reported that suffered stroke after operation19 and residual numbness and paraparesis were well documented. The capacities of the major systems in elderly patients is rather lower than that of younger patients, and their physical condition is usually weakened by surgery and long-term bed-stay. Thus, as to elderly patients with neurologic deficits and recently underwent intradural surgery, complications including hypostatic pneumonia, pulmonary-related heart failure, DVT, and hypopromelemia-related symptoms such as edema and delayed incision healing are more likely to happen. The occurrence of post-surgical complications in elderly patients is characterized by the “fuse” effect, which means that 1 complication is able to lead to several subsequent complications in other systems. In the present case, post-operative CF leakage exaggerated the perioperative complications that were resulted from the patient’s poor physical condition and surgery strike. She was required to stay in bed for 1 week due to cerebral fluid leakage caused by the dural incision, which led to hypostatic pneumonia and DVT formation. Moreover, the cerebral fluid leakage and hypoprotelemia worsened her inner osmotic pressure, which subsequently induced the hydrothorax and swelling in her lower extremities. Thus, effective suture of the dura matter to prevent serious leakage of cerebral fluid is necessary for elderly patients, which can effectively shorten the bed-rest duration and allow for early off-bed movement and can also help to prevent hypostatic pneumonia, DVT, and calcium loss.

4. Conclusions
We presented a rare case of a patient with OSM, who was also the second oldest patient that underwent OSM resection. However,
due to the elderly patient’s poor physical condition, she developed post-surgical complications in multi-systems. Through this case, we summarized that performing intradural surgeries to elderly patients required well-designed surgical strategy, short surgery time, perfect closing of dura matter, and pre-operative physical preparation. Once the post-surgical complications occur, it is vital to “cut the fuse”. As long time bedrest is the main cause of several post-surgical complications in elderly patients, early off-bed movement is recommended. Along with the precise surgical resection of OSM, peri-operative treatment of elderly OSM patients also deserves greater attention.

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