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Case report

Coexistence of cervico-thoracic extradural en-plaque meningioma with multiple intracranial meningiomas

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ARTICLE INFO

Article history:

Received 15 January 2014

Accepted 31 July 2014

Available online 20 August 2014

Keywords:

Meningioma

En-plaque

Extradural

Spinal

Surgery

ABSTRACT

Meningioma is one of the most common tumors in the spinal cord. Extradural and en-plaque variety of meningioma occur less frequently. A 47-year-old woman is presented with radiculopathy signs. Magnetic resonance imaging revealed a lesion from C6 through T3 vertebral levels compressing the cord both anteriorly and posteriorly. Subtotally excision was performed and histopathologic signs showed transitional type of meningioma (WHO Grade 1). Post operatively, she had good neurological recovery. Intraoperative findings point out that the en-plaque meningioma was pure extradural. Twelve cases of pure extradural en-plaque meningioma have been reported in the literature. Besides, to the best of our knowledge coexistence of “en plaque” spinal epidural meningioma with meningiomas in cranial cavity has not been reported. Complete resection is mandatory to prevent recurrence. Moreover, it is considerably difficult to remove the parts of tumor over anterior of the dura without complication.

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1. Introduction

While such malignities as metastasis and lymphoma prevail in spinal extradural region, meningiomas are mostly confronted as extramedullary neoplasms. Intraspinous meningiomas constitute 25–46% of primary spinal cord tumors whereas

extradural spinal meningiomas constitute 2.6–10% of spinal meningiomas [1–5]. It is believed that extradural spinal meningiomas are caused by nerve roots, where duramaters are thinner; thus they spread to extradural distance easily. What is assumed for extradural localization is migration of syncytial cells to extradural space or invasion of dura by these cells [6–8]. Meningioma usually occurs as the round type,

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Table 1 – Reported cases of spinal extradural en-plaque meningiomas.

No. of cases	Authors	Age/sex	Location
1	Achari et al., 2000 [9]	40/M	C3-4
2	Calogero and Moossy 1972 [10]	33/M	T7
3	Calogero and Moossy, 1972 [10]	54/M	L3
4	Calogero and Moossy, 1972 [10]	40/F	T4
5	Calogero and Moossy, 1972 [10]	28/F	T9-11
6	Gamache et al., 2001 [11]	63/F	C3-6
7	Kyoushima et al., 1987 [12]	26/F	T2-4
8	Messori et al., 2002 [13]	14/F	C5-7
9	Yamada et al., 2007 [14]	22/F	C1-5
10	D'Amico et al., 2012 [15]	74/F	C3-T1
11	Mariniello et al., 2012 [16]	74/F	C4-T1
12	Tuli et al., 2012 [17]	42/F	T4-6
13	Present case	47/F	C6-T3

whereas the en-plaque type grows in a sheet-like form along the dura mater. Extradural and en-plaque meningioma type is so rare such that, to the best of our knowledge, twelve cases of pure extradural en-plaque meningioma have been reported [9–17] (Table 1). In our article, we present pure extradural en-plaque meningioma located in cervico-thoracic region with meningiomas in cranial cavity of an adult female patient.

2. Case

A 47-year-old woman consulted our hospital with complaint of neck and left arm pain, numbness and strength loss in the left arm. Her clinical history revealed a tumor excision from the left parietal region with craniotomy 10 years ago and the pathology of the lesion was evaluated as grade 1 meningioma. At the time of the diagnosis, multiple intracranial of small lesions were detected but no additional treatment was taken. During follow-ups, the size of the lesions remained constant. Additionally, it has been reported that she had operations due to lumbar disc herniation, goitre and a gynaecological disease. No feature was detected in her family history. In her neurological examination, motor strength 3/5 in the left fore arm extensor muscle, C-7,8 hypoesthesia in the left side and loss of triceps reflex were detected. Cervical spinal MRI showed

a large mass lesion compressing the cord anteriorly and posteriorly with a craniocaudal extension of 7.3 cm at the level of C6-T3 vertebrae and 1.1 cm in its thickest part. Tumor was widening and extending through T1 foramina to encroach the paravertebral tissue. It was isointense to the spinal cord on T1 and hyperintense on T2 weighted images with showing intense homogeneous contrast enhancement (Fig. 1A–D). On thoracic spinal MR examination, another lesion of 13 mm × 8 mm in accord with meningioma at the level of T9 vertebra was detected (Fig. 2A and B). Cranial MR examination showed a total of 7 more extra-axial mass lesions in accord with meningioma, and post operative changes at the left parietal region due to the cranial operation (Fig. 2C). Preoperative preparation of the patient was made and the operation was carried out. Although anterior approach with vertebrectomy could be used to remove the ventral part of the tumor, we preferred posterior approach. In prone position, laminectomy was carried out between C6-T3 and facetectomy from left side to T1 was carried out, then mass lesion was seen within epidural space. The tumor was dissected from the dura dorsally, and then gently teased away from the ventral dura using a hook (Fig. 3). Its posterior part was removed completely; and the parts extending to anterior of the dura were excised subtotally by leaving some residue in anterior and medium regions. Dura was opened and intradural region was controlled and no intradural component of the tumor was detected, it was accepted as pure extradural. Posterior stabilization was ensured by using bilateral C6 lateral mass, C7-T3 transpedicular screw. In postoperative examination, it was observed that her pain went away, strength loss became normal and other findings remained unchanged. Cervical MRI examination was carried out at a postoperative early stage (24th hour), it was observed that spinal cord was prominently relieved and some residual tumor tissue seen in anterior or dura (Fig. 4). As a result of histopathological evaluation of removed tissues, it was diagnosed as a transitional type of meningioma (WHO Grade 1) (Fig. 5). We planned adjuvant radiotherapy for the residual tumor.

3. Discussion

According to the literature, 85% of spinal meningiomas are intradural located, 7% of them are intra-extradural located, and 8% of them are extradural located [3,5]. No tumor was detected in control of intradural space of our patient and it was

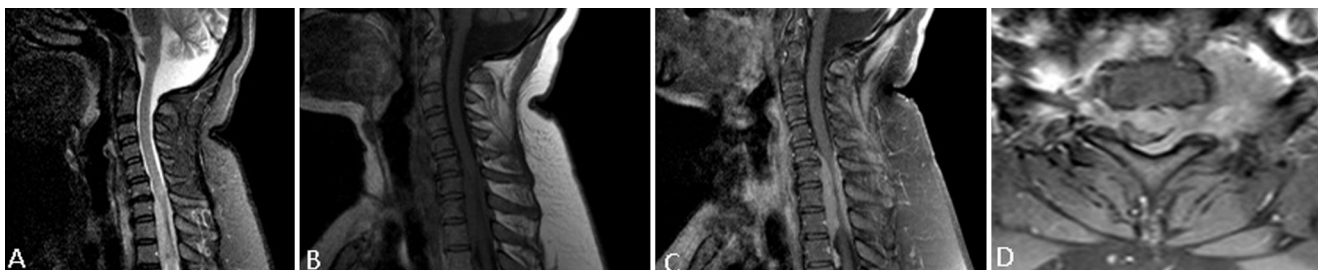


Fig. 1 – Sagittal T2-weighted (A) and sagittal T1-weighted (B) MR images show the extradural mass lesion that was hyperintense on T2-(A) and isointense on T1-(B) weighted images. The lesion shows intense and homogeneous enhancement on postcontrast T1-weighted sagittal (C) and axial (D) image.

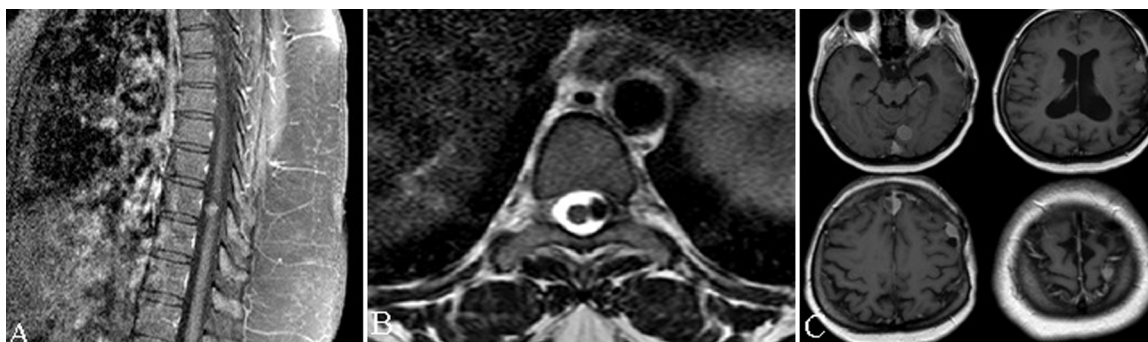


Fig. 2 – Thoracic (A, B) and cranial multiple lesions (C) are shown.

accepted as pure extradural. In patients diagnosed with extradural mass lesion, metastatic disease should be eliminated. Other most frequent pathologies are schwannoma, neurofibroma, chordoma, synovial cyst, infectious process and meningiomas [18]. As our patient was operated due to cranial meningioma previously and she had multiple craniospinal mass lesions in accord with meningioma radiologically, firstly we thought of meningioma as prediagnosis. Extradural meningiomas are most frequently observed in thoracic region; then in cervical region [7]. In this review, it is shown that, 4 patients have cervical, 5 patients have thoracic and 1 patient has lumbar originated tumors. In our patient, the tumor was extending from lower cervical to upper thoracic as the other 2 patients which were shown on the table. Extradural and en-plaque meningiomas had tendency to extend across a few vertebrae corpora on dura and they have difficulties in their treatments due to total extraction difficulty.

While in some cases spinal en-plaque meningiomas have intradural and extradural components, most of them are located intradurally. Generally, they are not adherent to the dura, but they have tendency to invade the adjacent nerve root or they appear to be adherent to it [17]. In our patient, no intradural infiltration was detected microscopically at the operation.

While frequency of spinal meningiomas is higher in young women, some publications indicate that extradural meningiomas are seen more frequently in children and men

[9,10,13,19,20]. When we evaluate the 13 patients with the extradural en-plaque meningioma, 10 (76.9%) of them were female and 3 (23.1%) of them were male at the average age of 42.8. While none of these spinal cases has indicated an additional meningioma occurrence, in our case it shows multiple intracranially meningiomas.

While patients mostly apply with progressive myelopathy findings; the first symptom may rarely be radiculopathy [11,21]. Onset symptom of our patient was radiculopathy caused by radicular compression depending on intervertebral foraminal extension.

MRI has replaced myelography and CT in diagnosis of these patients and patient follow up. Meningiomas are observed as isointense with spinal cord in T1 weighted images, and they are observed as isointense or light hyperintense in T2 weighted images, and intense contrast involvement is expected [13]. While calcification is seen in 1-4.6% of all spinal meningiomas, this rate is more frequent in en-plaque type [13,22]. No calcification was detected in our patient.

Despite notifications with regard to meningioma after trauma, the relation of trauma and meningioma is debated

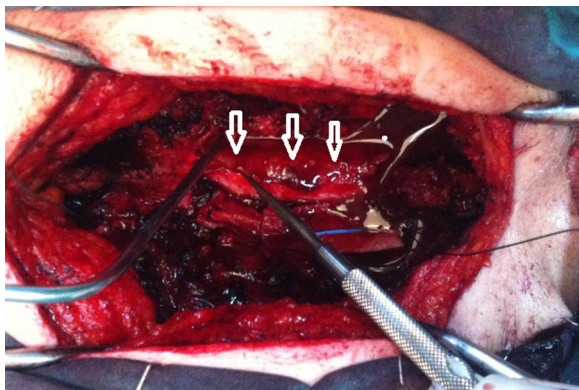


Fig. 3 – Intraoperative sight of the extradural en-plaque mass lesion (arrows) laying over the dura.

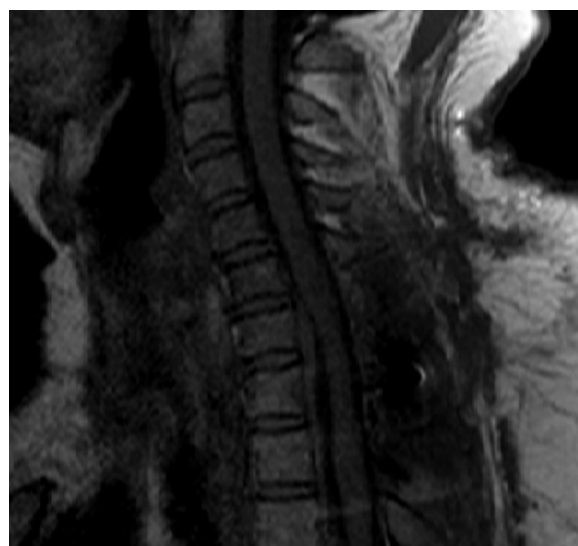


Fig. 4 – Subtotal removal of the tumor and decompression of the spinal cord are shown on sagittal T1 weighted image.

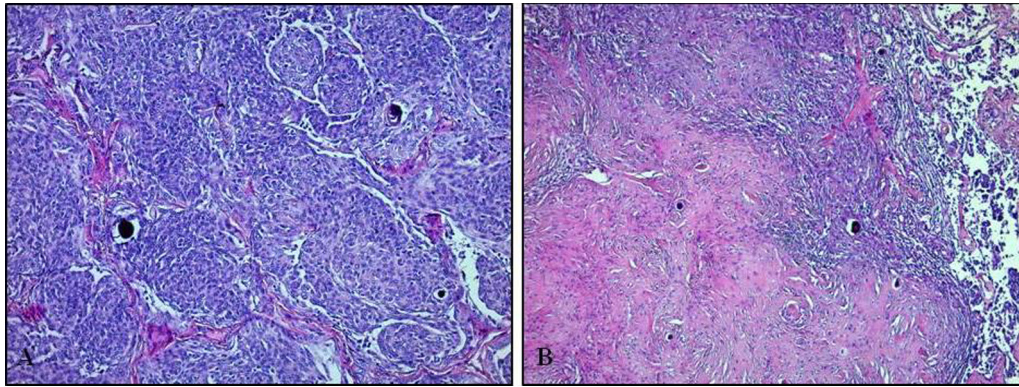


Fig. 5 – Histopathological appearances of the transitional meningioma (WHO Grade I). (A) The meningotheelial part of the tumor is composed of whorls, cords of neoplastic cells and psammoma bodies. (B) The fibrous component of the tumor is characterized by fibrous nodules and spindle cells (H&E; A,B, $\times 100$, $\times 50$).

and pathogenesis is still not clear [23,24]. When the history of our patient was examined, there was no trauma history.

Meningiomas very rarely metastasize into other organs. In the case of metastasis, it is believed that seeding is started by cerebrospinal fluid or venous sinus circulation. There is some evidence that surgical intervention may cause metastasis [25]. Metastatic parotid gland [26], lung, pleura [27], bone [28], lymph nodes [29] and skin [30] meningiomas have been reported in the literature.

In neurofibromatosis type II, cranial and spinal meningioma have been observed together. The relationship between isolated multiple meningiomas and neurofibromatosis is unsettled. In multiple cranial and spinal cases, supratentorial involvement is frequent in the absence of NF II [31,32]. In our patient, there exists mass lesion in accord with intracranial meningioma in 7 separate regions. Apart from that, there is a lesion in accord with meningioma at the level of T9 vertebra. Criteria of our patient do not meet NF II. However, in our study chromosomal analysis was not carried out.

In a study carried out with 54 intraspinal meningioma cases, 44.5% meningotheelial type was observed; at the same time transitional type was detected as 9% [33].

Surgical method is required for decompression of the cord by removing the tumor in treatment of extradural en-plaque meningioma. The important point in removing meningiomas is excision of the tumor tissue together with origin dura connection. However this is difficult in en-plaque meningiomas [9]. Additional treatment may not be necessary in most cases, in which the tumor can be removed completely. Moreover, it is considerably difficult to remove the parts of tumor extending over anterior of the dura without complication. Recurrence rate has been reported as 6–21% in spinal meningiomas [3,22,34]. Effects of age, subtotal excision, existence of calcification, multiplicity of lesions, anterior localization of the tumor on recurrence have been reported [2,3,33,35,36]. Furthermore, it has been indicated that if complete resection is ensured, prognosis will not change [3]. It is reported that, in en-plaque type, recurrence rate is higher than normal type, after 1 year 49.5% versus 7.9% and after 5 years 81.1% versus 20.4% [22].

In literature, there exists no information regarding adjuvant radiotherapy applied on spinal meningioma patients. In

long term follow up, the effect of radiation on recurrence is not known. Moreover, the enlargement of residual tissue is inevitable in this type of tumors, because of the difficulty to remove the whole part. In a recent study, irradiation was performed for a residual tumor and no retrogression was recorded at the 18 month follow up [16]. Some parts of the tumor of our patient could not be removed, recurrence may be expected in patient follow up; therefore, we preferred applying adjuvant radiotherapy.

Conflict of interest

None declared.

Acknowledgement and financial support

None declared.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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