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CASE REPORT

Symptomatic spinal cord metastasis from cerebral oligodendroglioma

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Abstract Spinal subarachnoid spread is not uncommon in brain oligodendrogliomas; on the other hand, symptomatic involvement of the spinal cord and cauda is very rare, with only 16 reported cases. We report the case of a 41-year-old man who underwent resection of a low-grade frontal oligodendroglioma 4 years previously. He was again observed because of bilateral sciatic pain followed by left leg paresis. A spine MRI showed an intramedullary T12–L1 tumor with root enhancement. At operation, an intramedullary anaplastic oligodendroglioma with left exophytic component was found and partially resected. Two weeks later, a large left frontoparietal anaplastic oligodendroglioma was diagnosed and completely resected. The patient was neurologically stable for 8 months and died 1 year after the spinal surgery because of diffuse brain and spinal leptomeningeal spread. The review of the reported cases shows that spinal symptomatic metastases can occur in both low-grade and anaplastic oligodendrogliomas, even many years after surgery of the primary tumor; however, they exceptionally occur as first clinical manifestation or as anaplastic progression. The spinal seeding represents a negative event leading to a short survival.

Keywords Brain glioma · Oligodendroglioma · Spinal metastasis · Cerebrospinal fluid spread

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Introduction

Brain gliomas may sometimes spread through the cerebrospinal fluid toward the spinal axis. The spinal seeding mainly occurs as diffuse microscopic subarachnoid spreading, whereas spinal cord localizations are very rare.

Spreading along the spinal canal has been reported mainly in patients with glioblastoma and anaplastic astrocytoma (up to 20% of cases with CSF positivity) [1]; it is also frequent for oligodendrogliomas (8.5 up to 14% of CSF microscopic seeding) [2, 3]. On the other hand, symptomatic involvement of the spinal cord from brain oligodendroglioma is very rare.

We report in this article a rare case of brain oligodendroglioma where a large symptomatic spinal cord metastasis was the first clinical manifestation of the anaplastic progression.

Case report

In March 2006, a 37-year-old man underwent surgical resection of a left frontal low-grade (WHO II) oligodendroglioma (Fig. 1). In July 2009, he underwent a second operation for tumor recurrence (WHO II oligodendroglioma with increased mitotic activity) and irradiation to the tumor field (60 Gy for 6 weeks at doses of 2 Gy for 30 days).

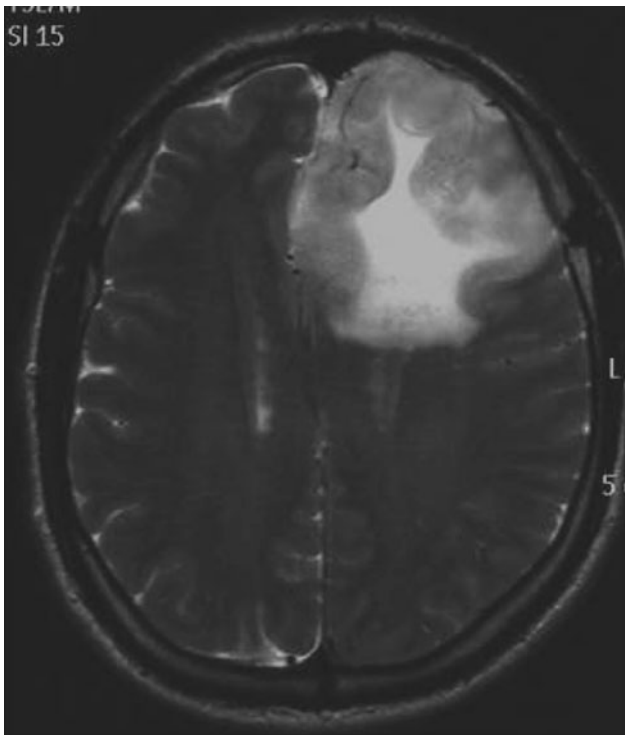

Fig. 1 Magnetic resonance of the brain before the first craniotomy (March 2006), axial T2W sequence: heterogeneous high-signal large tumor mass in the left frontal region, suggesting a low-grade glioma

Postoperatively, the patient was symptom free. Serial MRI studies showed no tumor recurrence.

However, in May 2010 the patient complained of sudden and intense left sciatic pain with distribution on the left S1 territory. The pain was continuous and not responsive to corticosteroids and other drugs. About 2 weeks later, weakness on the left leg with inability to perform flexion of the foot, less intense sciatic pain on the right leg and bladder retention also occurred.

Thus, the patient was again admitted to our neurosurgical clinic. Neurological examination revealed paralysis of the left anterior tibialis muscle, hypoesthesia of the left leg, and bilateral decrease of the knee and ankle reflexes.

MRI of the spine (Fig. 2a) showed an intramedullary tumor located in the posterior and left lateral part of the conus medullaris, with a greater diameter of about 3 cm, irregular margins and inhomogeneous contrast enhancement. Irregular linear enhancement of the contiguous lumbar nerve roots, mainly on the left side, and of the thoracic cord surface was also evident.

Therefore, the patient underwent surgery through a T12–L1 laminectomy. A reddish, markedly hemorrhagic intra-axial tumor with an exophytic component over the left aspect of the conus was exposed. A partial tumor resection, mainly of the exophytic portion, was possible, because of the diffuse infiltration and marked tumor bleeding.

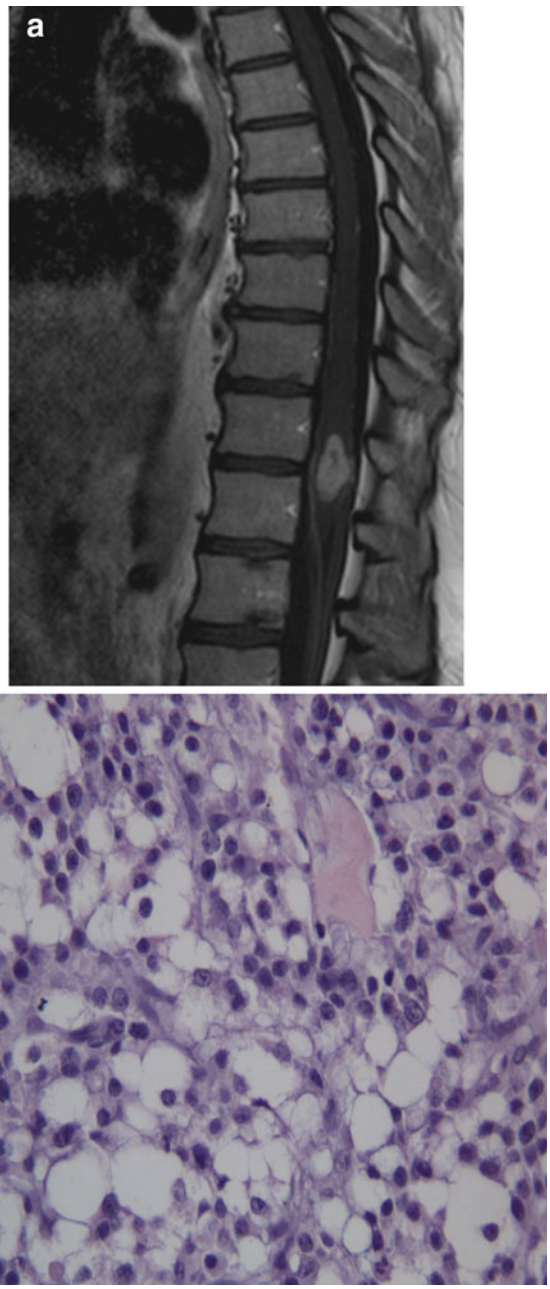

Fig. 2 **a** Spinal magnetic resonance (May 2010), gadolinium-enhanced sagittal T1W sequence: widening of the conus medullaris, due to a heteroplasic ovalar lesion with intense and heterogeneous enhancement. Thin linear enhancement of the thoracic cord surface is also evident. **b** Histological finding: neoplastic cells with round, hyperchromatic and mildly atypical nuclei and vacuolated cytoplasm, sometimes with clear perinuclear halo (anaplastic oligodendroglioma) (E/E ×40)

The CSF sample, obtained from the surgical field before the tumor resection, was positive for oligodendroglial tumor cells. Histological examination of the surgical specimen was in favor of an anaplastic (WHO III) oligodendroglioma with cytological aspects of aggressiveness

and areas of hypercellularity and pleomorphism (Ki-67 LI 30%) (Fig. 2b).

In the postoperative course, weakness and numbness in the lower limbs worsened to a moderate paraparesis, more marked on the left side. MRI of the lumbosacral spine confirmed the presence of residual intramedullary tumor with enhancement of the nerve roots. Thus, radiotherapy to the thoracolumbar region (T9–L3) with 45 Gy was started 2 weeks after the operation.

However, 10 days after the onset of radiotherapy, the patient presented a right-sided partial motor seizure. Brain MRI (Fig. 3) revealed a large (>4 cm) tumor mass in the left prerolandic region, with prevalent exophytic component, homogeneous contrast enhancement and mass effect. Peripheral enhancement of the surgical field of the previous operation was also evident, suggesting tumor progression.

Thus, the mainly exophytic tumor mass was removed through a left frontoparietal craniotomy. The histological examination of the surgical specimen confirmed the diagnosis of anaplastic (WHO III) oligodendroglioma.

Postoperatively, transient dysphasia occurred for about 10 days. One month later, brain MRI showed no residual tumor in the left frontoparietal region and stable lesion of the left anterior frontal region. Thus, the radiotherapy to the spine was completed.

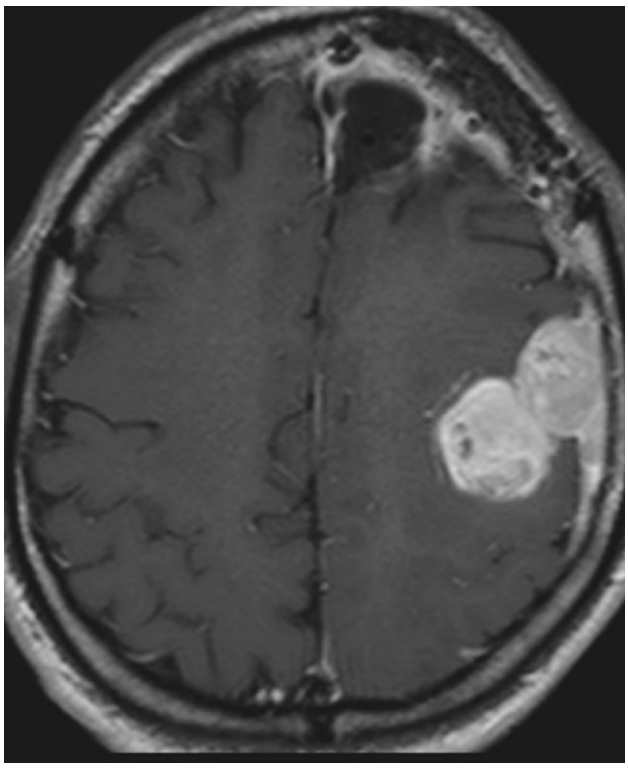

Fig. 3 Brain magnetic resonance (June 2010), gadolinium-enhanced axial T1W sequence: in the left prerolandic region, the presence of a large neoplastic intracerebral mass contiguous with the dura mater is seen; both the tumor and the dura show intense and heterogeneous enhancement. In the left frontal region, at the level of the previous surgical site, a linear peripheral enhancement is evident

The patient was neurologically stable, with severe paraparesis for about 8 months, and died 1 year after the spinal surgery because of diffuse brain and spinal leptomeningeal spread.

Discussion

The possible extensive spread of oligodendrogliomas through the CSF in the spinal subarachnoid spaces has been known since 80 years [4, 5]. However, metastatic involvement of the spinal cord and nerve roots has been reported mainly as microscopic findings; on the other hand, macroscopic and symptomatic spinal metastases are very rare, with only 16 reported cases (Table 1) [6–18].

Because of their exceptional occurrence, the real incidence of these macroscopic symptomatic spinal localizations has not been defined. Arseni et al. [11] found 2 cases among 170 oligodendroglial tumors (1.2%), and Wallner et al. [13] 1 among 40 cases (5%).

The 17 reported cases (including our own) (Table 1) show a wide age range (6.5–73 years, median age 37 years). The site of the primary brain oligodendroglioma was mainly in the cerebral lobes, as it is usual for these tumors. However, in 3 of the 17 cases (18%), the tumor was located in the lateral or third ventricle; this rate is significantly higher than that reported for the overall oligodendrogliomas [19]. Besides, in our case the tumor mass was mostly exophytic in the subarachnoid space. These peculiar tumor locations may account for the spinal spread.

From the histological viewpoint, the reported cases show a similar rate of low-grade and anaplastic oligodendrogliomas. This confirms that spinal spread depends more on the tumor location near the CSF pathways than on the histological grade.

The time interval between surgery of the brain tumor and diagnosis of the spinal metastasis ranges from 3 months to 7 years (average 25.5 months). It is slightly longer for low-grade oligodendrogliomas (30 months) than for anaplastic tumors (21 months). In the case reported by Natale et al. [16], the symptoms of cauda equina involvement were the first clinical manifestations of the intracranial anaplastic oligodendroglioma. In our case, the spinal symptoms first occurred on progression from low-grade to anaplastic oligodendroglioma with no brain symptoms.

Spinal metastases are more often diffuse or involve the cord surface and roots over a short spinal segment (2–4 metameres). However, in three cases the spinal metastasis was intramedullary [12, 15, 18]. In our case, a mostly intramedullary lumbar tumor with exophytic component and root infiltration was found at operation.

Table 1 Data of reported cases of symptomatic spinal metastases from cerebral oligodendrogliomas

Authors	Age/sex	Site of the primary tumor	Spinal metastasis	Interval	Histology	Survival
Beck and Russell [6]	29/M	L. lateral ventricle	Diffuse	3 years	LGO	–
	42/M	R. frontal	Diffuse	Synchronous	LGO	–
	6.5/M	R. frontal	Diffuse	3 months	LGO	–
Trowbridge and French [7]	38/M	R. temporoparietal	Diffuse	6 years	AO	–
Strang and Nordenstam [8]	30/M	R. frontal	Cauda	5 years	AO	–
Reggiani et al. [9]	43/F	L. frontal	T3–T5	7 years	LGO	–
Voldby [10]	27/F	R. frontal	Diffuse	6 months	AO	–
Arseni et al. [11]	11/M	3rd ventricle	C1–C5, extramedullary	2 years	LGO	–
	39/M	R. temporal	T1–T3	15 months	LGO	1 month
Van Velthoven et al. [12]	13/F	R. parietal	T6–T8, intramedullary	8 months	LGO	2 months
Wallner et al. [13]	40/M	L. temporal	T11, extramedullary	6 years	LGO	2 months
Ng et al. [14]	61/F	L. frontal	C7-conus (diffuse)	20 months	AO	4 months
McBryde et al. [15]	49/M	R. parietal	C7–T2, intramedullary	14 months	LGO	Alive
Natale et al. [16]	67/F	L. lateral ventricle	Cauda	Synchronous	AO	8 months
Ozisik et al. [17]	40/M	R. frontal	Diffuse	1 year	AO	1 year
Oshiro et al. [18]	73/F	Cerebellar	C2–C4, intramedullary	18 months	AO	–
Present report	40/M	L. frontoparietal	T12–L1, intramedullary and nerve roots	Synchronous	AO	1 year

LGO low-grade oligodendroglioma (WHO II), *AO* anaplastic oligodendroglioma (WHO III)

Surgery of spinal metastases from brain oligodendrogliomas may be advisable only for cases with localized tumors and no diffuse root involvement. The aim of the operation is to decompress the cord and cauda equina roots. However, the resection is partial due to the extensive infiltration of the nervous structures.

Radiotherapy to the spinal axis has been performed in several reported cases [12, 16–18], including ours, with a dose of 45 Gy. The results of the radiation therapy are difficult to define, due to the short survival. We agree that irradiation should be performed in patients with localized spinal metastasis, good Karnofsky performance status and sufficient life expectancy. Prophylactic spinal radiation therapy is not justified, due to the rare occurrence of these symptomatic metastases.

The occurrence of spinal metastases is a negative event in the history of oligodendroglial tumors. In all seven cases, which report the survival time (Table 1), it ranges from 1 month to 1 year.

Spinal metastases occur as a result of dissemination from the brain superficial or ventricular oligodendrogliomas through the subarachnoid spaces. Because most cases occurred after surgery of the brain tumor, surgical manipulation may play a role. In exceptional intramedullary metastases, spread through the central canal [20] or, more likely, through the perivascular Virchow-Robin spaces of the spinal cord [21] has been suggested.

Conclusion

Brain oligodendroglioma may exceptionally spread into the spinal canal. The symptoms of spinal involvement may occur in both low-grade and anaplastic tumors even several years after the treatment of the primary tumor; however, they may also appear before brain symptoms or at anaplastic progression. Thus, the occurrence of a spinal cord or nerve root syndrome in a patient with oligodendroglioma even many years after the initial surgery must suggest a spinal metastastic localization.

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