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Intracranial Metastases of Intramedullary Spinal Cord Low-Grade Astrocytoma

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Abstract

This paper reports a case of intramedullary spinal cord low-grade (LG) astrocytoma that developed brain metastases after 21 months. A 6-year-old child presented with lower spine pain and falls during daily activity. A spinal cord mass was detected using spinal magnetic resonance imaging (MRI), and brain MRI was normal. The spinal lesion was partially resected, and pathological findings revealed LG astrocytoma (WHO Grade II). The patient underwent thoracolumbar radiotherapy. He returned 21 months following initial admission with symptoms of nausea, vomiting, headaches, and seizure. Brain MRI revealed multiple intracranial masses at the posterior fossa, left lateral ventricle, cerebellopontine angles, and left Meckel cave. A recurrent lesion was detected in the thoracic and lumbar regions of the spinal cord and in the cauda equina. The patient underwent chemotherapy. This rare case warns practitioners to monitor closely the cases of spinal cord astrocytoma that are diagnosed as LG tumors based on histology.

Keywords: *Intracranial metastases, low-grade astrocytoma, pediatric, spinal cord tumor*

Introduction

Spinal cord astrocytoma is a rare neoplasm in the pediatric population.[1] The previous literature has reported that its prevalence is <1% of all primary neoplasms of the central nervous system[2-4] and that it comprises 6%–8% of all primary spinal cord tumors.[5-7] Brain metastasis of a primary spinal cord astrocytoma has rarely been reported, and most of the reported cases have been caused by high-grade astrocytoma.[8,9] However, very few cases of intracranial metastasis of low-grade (LG) spinal cord astrocytoma have been reported in pediatrics.[1] Abel *et al*. have reported a spinal cord pilocytic astrocytoma that disseminated to the cerebral subarachnoid spaces of a 2-year-old boy.[10] Jang *et al*. reported brain metastasis of an intramedullary LG astrocytoma in a 45-year-old patient. This study concluded that the LG spinal cord astrocytoma might spread to intracranial structures without malignant transformation.[4] Ryu *et al*. reported two malignant transformations of LG spinal cord astrocytoma among 12 patients.[2] Yamagami *et al*. reported a case of a 44-year-old patient with LG astrocytoma that metastasized to the brain after 6 years.[11]

This case report presents intracranial metastasis of LG spinal cord astrocytoma in a child. The purpose of this paper is to provide further evidence for practitioners about the metastasis of initially diagnosed LG spinal cord astrocytoma to the brain specifically in pediatrics.

Case Report

A 6-year-old boy presented with a 1-month history of lower back pain and left crural monoplegia. Spinal magnetic resonance imaging (MRI) revealed an intramedullary spinal cord tumor at the Level T8–T12. Partial resection of the tumor was performed, and the pathological findings of the resected tumor were consistent with LG astrocytoma (WHO grade II). A histological examination of the tumor showed tumoral tissue composed of neoplastic astrocytes with increased cellularity, mild atypia, and low mitotic activity. No vascular endothelial proliferation or necrosis was present [Figure 1].

A total dose of 45 Gy was delivered to the spine over 6 weeks. After 2 months, the patient returned with lumbar pain and

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paraplegia due to a spinal cord abscess [Figure 2]. The spinal cord abscess was removed in the second operation. Paraplegia and urinary/fecal incontinence appeared after surgery. The patient visited as an outpatient several times due to recurrent urinary infections caused by a neurogenic bladder, which occurred before the diagnosis of brain metastasis.

Twenty-one months after the first visit, the patient was hospitalized due to seizures and severe headaches. A brain MRI revealed multiple enhancing extra-axial masses at the posterior fossa, left lateral ventricle, cerebellopontine angles, and left Meckel cave, which was compatible with intracranial metastasis [Figure 3]. An elongated, expansile, partially cystic mass was detected in the lower thoracic and lumbar regions of the spinal cord, which extended to the cauda equine [Figure 4]. An assessment for neurofibromatosis Type 1 was negative in this case. The patient underwent the chemotherapy treatment with CCNU, vincristine, and cisplatin. Six months following the diagnosis of cerebral metastases, the patient died due to sepsis caused by the urinary tract infection.

Discussion

This paper reports a case of intracranial metastasis of intramedullary LG astrocytoma in a child. Most cases of intracranial metastasis of spinal cord astrocytoma are related to high-grade tumors and intracranial metastasis of spinal cord LG astrocytoma is a rare phenomenon.[4,10,12] The mechanism of intracranial metastasis of intramedullary spinal cord LG astrocytoma is not well known.[10,13] Some previous researchers have hypothesized that metastasis of LG astrocytoma can develop due to resection and manipulation.[14] However, in a review article, Abel *et al*. reported that none of the current studies have proved a correlation between the resection of a LG tumor and its metastasis.[10] Malignant transformation has also been reported as a reason for the brain metastasis of spinal cord LG glioma.[15,16] However, there is some evidence of brain metastasis of intramedullary LG astrocytoma without malignant transformation.[4,10] Malignant transformation of LG glioma in children is very unusual compared its occurrence in adults. Irradiation and genetic disorders such as neurofibromatosis-1 were proposed as predisposing factors for malignant transformation of LG glioma.[16,17] In our patient, radiotherapy had been administered after partial resection of the spinal cord tumor. Intracranial metastasis of the recurrent spinal cord tumor occurred approximately 2 years following the diagnosis of the primary tumor. Malignant transformation is thought to be a possible cause of metastasis in our patient. It was impossible to perform a biopsy on the patient's recurrent spinal cord tumor due to his condition.

Another hypothesis about our patient is a sampling error in a mixed tumor. In other words, the pathological examination may have been performed on a portion of the

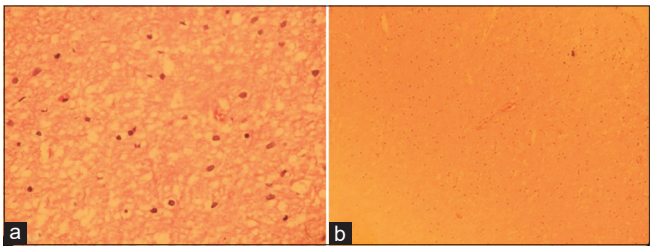

Figure 1: Histological study of the spinal cord tumor (a: H and E, ×400 and b: H and E, ×100) revealed neoplastic astrocytes with slightly pleomorphic enlarged nuclei and no visible cytoplasm, set in a loose fibrillary glial matrix consistent with low-grade astrocytoma

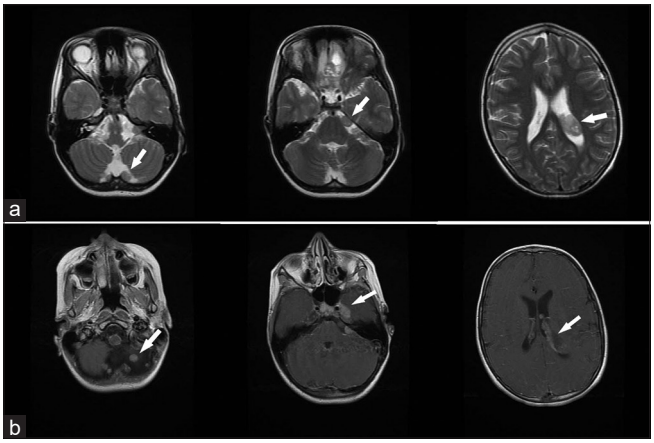

Figure 3: T2-weighted magnetic resonance image (a) and T1-weighted magnetic resonance image with contrast (b) showing multiple extra-axial enhancing masses at the posterior fossa, left lateral ventricle, cerebellopontine angles, and left Meckel cave

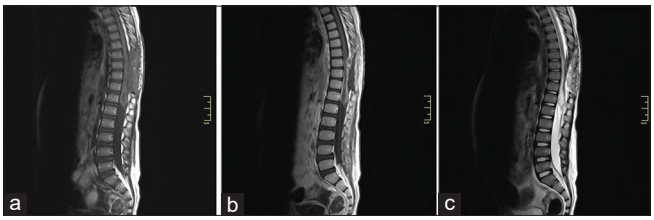

Figure 2: T1-weighted magnetic resonance image with/without contrast (a and b), and T2-weighted magnetic resonance image (c) showing spinal cord abscess appeared following the first operation

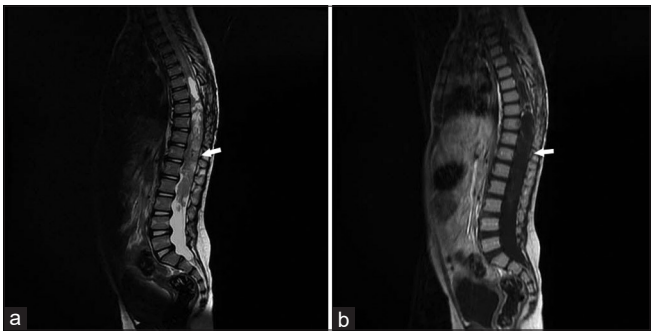

Figure 4: T2-weighted magnetic resonance image (a) and T1-weighted magnetic resonance image with contrast (b) showing an elongated, expansile, partially cystic mass detected in the lower thoracic and lumbar regions of the spinal cord, which extended to the cauda equina

primary tumor that contained only the LG tumor and the more invasive part of the tumor was not biopsied. Some studies have reported that histological techniques are probably insufficient to predict the future behavior of a LG glioma. Using biological markers and molecular genetics as parallel techniques is necessary to predict the tumor's outcomes and behaviors more accurately.[10,13] Ryu *et al*. concluded that although the pathological grade of the tumor is the most important prognostic factor, the biological behavior of the tumor is not always compatible with the pathological findings.[1,2] A treatment plan should therefore be formulated based on imaging so as to achieve a better therapy.[2] However, there are insufficient radiological criteria to distinguish between benign and malignant tumors based on imaging techniques.

Inconsistent information has been reported on therapy for intramedullary spinal cord astrocytoma, particularly in children.[2,6,7,17-20] Gross total resection (if possible) is reported as the principal treatment for LG spinal cord astrocytoma. Adjuvant radiation therapy is used for partially resected tumors.[20-22] However, several studies have reported high levels of morbidity due to more aggressive therapeutic approaches to LG glioma, which have an extreme influence on quality of life.[19,23] It was the case in our patient, as he had also a high level of postoperative morbidity. Aggressive treatment has therefore only been suggested in specific cases.[19] The role of radiotherapy and chemotherapy for spinal cord astrocytoma is unclear,[22] and these therapies may influence the tumor's biological behavior and increase the risk of malignant transformation.[16]

This case report warns clinicians regarding the treatment plan of LG spinal cord tumors. The patient should be carefully monitored, and particular attention should be paid to correlating the imaging of the tumor with pathological confirmation.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal patient identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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