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Case Report

The Role of the Surgery in the Case of an Intramedullary Spinal Cord Metastasis as a Primary Presentation of a Malignant Disease

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ABSTRACT

Introduction: Spinal cord metastases represent a small proportion of intramedullary tumors. The occurrence of such lesion over the course of malignant disease is usually a predictor for shortened life expectancy and is often associated with severe neurological deficits. Treatment options include microsurgical excision, radio-, chemo- or palliative therapy. Despite these possibilities the optimal management of patients with intramedullary spinal cord metastases (ISCM) is difficult due to the wide variety of clinical situations and the lack of controlled studies on the results of different therapeutic options.

Materials: We are presenting a case of a 68-year-old male with gradually increasing spinal neurological deficit – axial low back pain, numbness and muscle weakness in both legs and gait disturbance. Arterial hypertension was pointed as the only comorbidity of the patient. The MRI showed an intramedullary solitary lesion in the lumbar intumescence at the level of Th12 and L1 vertebra.

Results: The patient underwent surgery and the tumor was totally excised. The symptoms improved dramatically. The histology evidence a metastasis from a lung adenocarcinoma. Six months later he presented with severe back pain, progression of the paresis and urinary retention. Tumour recurrence was detected by MRI. After careful decision making and taking into consideration the risk of permanent neurological deficit a reoperation was performed. The lesion was gross-totally removed with improvement in functions and no neurological deterioration registered in the postoperative period.

Conclusion: It is possibly the symptoms from an intramedullary metastatic lesion to precede the detection of the primary tumour. The low frequency of occurrence and the absence of a manifested and diagnosed primary malignant process should not stop us thinking in the direction of metastasis. Surgical resection with function preservation is highly aimed. Operation and even reoperation in some cases is acceptable.

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Introduction

Spinal cord metastases represent a small proportion of intramedullary tumors. They are estimated to affect between 0.5% and 2.0% of all patients with cancer, representing only 8.5% of all CNS metastases and comprising 1–3% of all intramedullary spinal cord tumours (ISCT) [1–4]. Compared with neoplastic epidural compression, intramedullary spinal cord metastasis (ISCM) are diagnosed much less frequently, with an incidence approximately one-sixteenth as high [1, 2, 5]. The occurrence of such pathology over the course of malignant disease is usually a predictor for shortened life expectancy and is often associated

with severe neurological deficits. It can also suggest an advanced form of cancer and a biologically aggressive disease.

ISCT can arise as metastatic deposits within the spinal cord parenchyma from any type of primary cancer, but close to 50% of them result from primary lung cancer, with small cell carcinoma being the most common, and the remainder originating from primary cancers of the breast, colon, kidney, from melanomas and lymphomas [6]. The most frequent pathway for dissemination is the arterial route, direct invasion through the spinal roots or leptomeninges can also occur and retrograde spread through the Batson plexus during the Valsalva maneuver has also been

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suggested [7]. Based on the data in the literature the cervical segment of the cord is the most and the lumbar is the least affected, with thoracic segment staying in between [8-10]. Symptoms at presentation are usually pain and/or weakness. They are often followed by sensory level and sphincter disturbance. Rapid deterioration, within a period of days to weeks, with progression to a cord hemisection syndrome (Brown-Sequard) or full cord transection is possible.

Case Description

A 68-year-old male presenting with a signs of spinal cord compression - axial low back pain, numbness and muscle weakness in both legs and gait disturbance. The symptoms appeared 10 days ago and increased gradually in time. Arterial hypertension was pointed as the only comorbidity of the patient. The neurological examination revealed mild flaccid paresis, decreased reflexes and hypesthesia in the lower limbs. The patient underwent MRI of the spine, showing single solitary intramedullary lesion, located in the lumbar intumescence at the level of Th12 and L1 vertebrae (Figure 1). The lack of history for primary cancer made us assume that the type of the tumour is an ependymoma. This and the progression of the neurological symptoms were perceived as indications for surgery.



Figure 1: Sagittal and axial contrast-enhanced MRI images, showing intramedullary tumour at the level of Th12 and L1 vertebrae.



Figure 2: Intraoperative ultrasound view of the tumour.

A standard operative approach was used – prone position, median skin incision, bilateral paraspinal muscle stripping, laminectomy of Th12 and L1 vertebrae. Then ultrasound was used for verifying the exact location of the lesion (Figure 2), followed by opening the dura (Figure 3) and small myelotomy right over the tumor projection (Figure 4). After careful dissection the tumor was gross totally removed (Figure 5) with no changes in the intraoperative monitoring parameters. No new

neurological deficit was observed after the surgery. The muscle strength increase, and the pain diminish in the postoperative period, the sensory disturbance remains unchanged. The histological diagnosis, including immunohistochemical analysis substantiate non-small-cell lung adenocarcinoma. The patient was discharged and referred for subsequent treatment in oncology department. Three months after the surgery he underwent spine MRI for disease follow up. There were no symptoms present, however a small tumor relapse [with a diameter 0,85 cm] was evidenced (Figure 6).

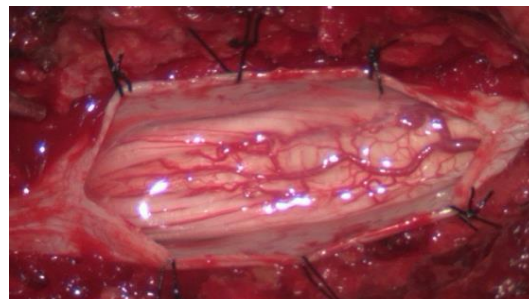


Figure 3: Intraoperative image after opening the dura, showing the spinal cord at the level of the lumbar intumescence.



Figure 4: Intraoperative image showing the site of the myelotomy. The incision is located between the pial veins of the coronal venous plexus.

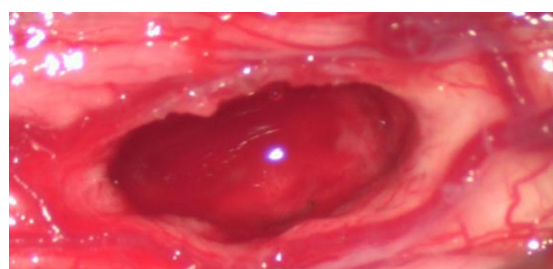


Figure 5: Intraoperative image after removal of the lesion, showing the tumor cavity in the spinal cord.

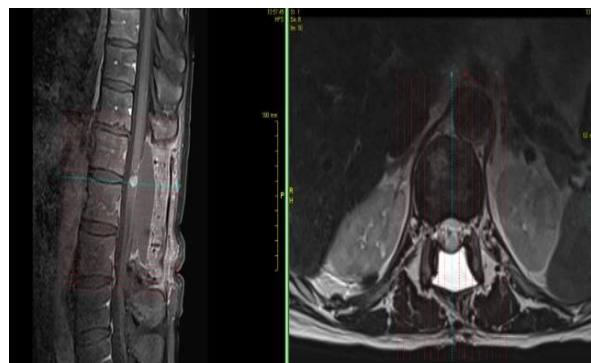


Figure 6: Sagittal and axial MRI images, performed 3 months after the surgery, showing small relapse of the lesion.

Operative treatment was offered, but declined, mainly due to the lack of clinical manifestation. The prime treatment method was chemotherapy and it was relied on its effect. After another three months or totally six after the surgery the patient was admitted with severe back pain, paraparesis, causing inability of standing and walking and newly emerged urine retention. The conducted MRI showed distinct increase in tumor's size (Figures 7 & 8). After careful discussion, taking into account the patient's wish for carrying out the surgery, and on the other hand, the potential risk for permanent neurological morbidity, a decision for performing a reoperation was made. The same surgical approach was undertaken, a lot of adhesions and a retained epidural CSF collection were present. The lesion was located mainly in the tumor cavity, which made possible its complete removal. The patient recovered well, the paraparesis improved, the pain was gone and there was no evidence for urine retention. He was discharged, fully able to walk on his own and referred to oncology for continuing the treatment.

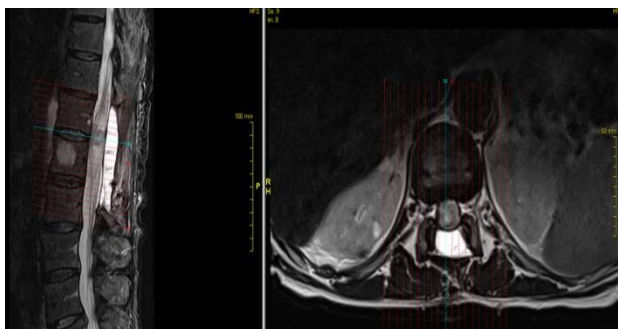


Figure 7: Sagittal and axial MRI images, performed 6 months after the surgery, showing clear increase in the size of the lesion.

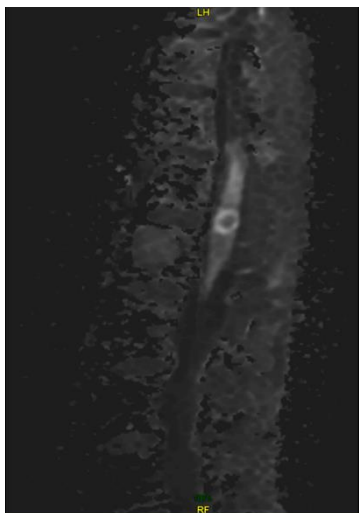


Figure 8: Sagittal diffusion-weighted MRI, performed 6 months after the surgery, showing clear increase in the size of the lesion.

Discussion

The proper diagnosis of ISCT is essential to provide the best treatment and obtain the optimal outcome. Intramedullary secondary lesions usually occur in the context of a well-established diagnosis of cancer. However, in some patients, the manifestation of symptoms can be misleading, or the primary disease can be occult and then the diagnosis can be difficult. Sung *et al.* performed a retrospective review of 301 patients and found that in 26% of the cases the detection of the ISCM led

to the ultimate diagnosis of the systemic cancer [1]. By the majority of these patients the definitive histological diagnosis of the ISCM and the primary disease resulted from a surgical biopsy or resection [11-23].

There are some signs that can suggest a secondary lesion. While slow progression of the symptoms (mainly mild pain and neurological deficit) is typical for primary spinal cord tumours, one rapid deterioration speaks for a metastatic lesion. In addition, muscular atrophy and fasciculation are being more frequently observed in primary intramedullary lesions when compared with ISCM [24, 25]. On the other hand, MRI can be very helpful – the metastases are usually solid with no cystic or haemorrhagic components (which can be present in the case of primary tumour) and there is distinct contrast enhancement with the so-called „rim” and „flame” signs. The first is a complete or partial, thin, peripheral rim of gadolinium enhancement more intense than the central enhancement of a noncystic/necrotic lesion and the second is an ill-defined flame-shaped region of gadolinium enhancement at the superior and/or inferior margin of an otherwise well-defined lesion [26].

And why a precise preoperative diagnosis is so important? Because of that depends the type of subsequent approach. The treatment modalities include surgery, radiation therapy (RT), chemotherapy (CT) or steroids. Surgery is the preferred choice in the case of primary intramedullary lesion. The goal is gross total removal, insofar as possible, without the occurrence of new neurological deficit. In the case of ependymoma (the most common spinal cord tumour in adults) or low-grade astrocytoma that would lead to cure of the disease, because relapse wouldn't occur or that could happen extremely rare. When it comes to high-grade astrocytoma or some other more malignant version of the primary tumour, where the lesion is infiltrative and borders with normal tissues are lost, the purpose of surgical treatment is to decompress the spinal cord and by that to prevent the progression of clinical symptoms and the onset of a persistent neurological mortality.

In the case of metastasis things are different. RT has been the treatment of choice since many years. Best responses are achieved in radiosensitive cases especially if implemented very early over the course of presentation [4]. Because of the risk for developing radiation myelitis a fractionated radiotherapy (FR) or stereotactic radiosurgery (SRS) are recently recommended. CT has been used in conjunction with RT or surgery in some chemotherapy-sensitive tumours. According to some authors, CT is not effective in the case of ISCM, because the drugs do not pass through the blood-spinal cord barrier (BSB) and have no further effect on survival [27-30].

Patients with rapidly progressive symptoms of cord compression and a high risk of rapid deterioration are suitable for treatment with high-dose steroids. This may decrease the pain and cause transient improvement in neurological conditions. Steroids suppress perifocal oedema and normalize the BSB, which decreases tumour size but does not influence survival [7].

The most common indications for performing surgery are the decompression of functional neural tissue and histological confirmation of the tumour. Currently, surgical approach is more precise and less invasive and thus allows total resection with no or acceptable morbidity rate. Considering the importance of total tumour resection in the case of cerebral metastases, one could postulate a similar importance when

surgically removing ISCM. Kalayci *et al.* demonstrated the length of survival in surgical patients being almost twice as long than the nonsurgical patients [4]. Connolly *et al.* also found that the survival correlated strongly with aggressiveness of therapy [12]. However, Wilson *et al.* as well as Watanabe *et al.* could not find a benefit from surgery concerning the length of survival in their respective series [23, 31]. Nevertheless, their patients had preoperative urinary retention, which is accompanied by a progressive neurological deficit and poor prognosis. Our patient also had that symptom before the second surgery.

Performing a reoperation for an intramedullary lesion is always a hazardous decision. The data in the literature reporting such action is very limited. Our considerations included the explicit wish of the patient for surgical intervention. He had severe pain and couldn't bear it. Even though the relapse of the lesion was bigger in size, there were less tumour blood vessels present and less bleeding, which made the dissection easier. Working only within the boundaries of the tumour, using careful microdissection technique, permitted gross total removal with no changes in the neurophysiological parameters and no appearance of neurological morbidity after the surgery.

Conclusion

It is possibly the symptoms from an intramedullary metastatic lesion to precede the detection of the primary tumour. The low frequency of occurrence and the absence of a manifested and diagnosed primary malignant process should not stop us thinking in the direction of metastasis. Surgical resection with function preservation is highly aimed. Operation and even reoperation in some cases is acceptable and can potentially result in significant improvement in the neurological function and quality of life.

Abbreviation

BSB: Blood-spinal cord barrier

ISCM: Intramedullary spinal cord metastasis

ISCT: Intramedullary spinal cord tumours

CNS: Central nervous system

CT: Chemotherapy

RF: Fractionated radiotherapy

RT: Radiation therapy

SRS: Stereotactic radiosurgery

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