CASE REPORT

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SPINAL ANGIOMATOUS MENINGIOMA IN THE THORACIC REGION: A CASE REPORT

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Abstract. *Meningiomas are divided into three subtypes according to the World Health Organization (WHO), which give rise to about 15 histological variants with varying degrees of malignancy. WHO grade I tumors are the most common, which are predominantly benign in nature and have a good prognosis after total surgical removal. Spinal meningiomas are one of the most common intradural, extramedullary tumors, accounting for about 20% of all lesions localized on the spinal cord and its envelope. Meningiomas arise from arachnoid cells and have a very close relationship with the dural meninges. They can be located in all places where there are arachnoid cells and cause neurological symptoms. Due to their slow-growing nature, symptoms manifest within a few months. In this report, we present an unusual case of a 44-year-old woman with spinal angiomatous meningioma presenting with the clinicopathology of inferior spastic paraplegia debuting at eight months, pelvic reservoir disorders, and decreased bowel motility, resulting in neurological paresis of the latter. The patient underwent two operations for hydrocephalus with normal pressure and subsequent infection of the ventricular catheter, which required its removal in the neurosurgical department and prolonged antibiotic treatment. The woman stayed for about three months in the intensive care unit with the clinical picture of meningitis and secondary superimposed pulmonary pneumonia. Magnetic resonance imaging of the thoracic region revealed a heterogeneous cystic formation over a wide area, and a tumor formation or arachnoiditis in the chronic phase was considered in the differential diagnosis. Surgical treatment with partial extirpation of the pathological formation (Simpsom grade III) was undertaken, and the histological diagnosis confirmed the presence of an angiomatous meningioma in the thoracic region.*

Key words: *spinal meningiomas, diagnosis and treatment of spinal tumors, histological outcome of spinal meningiomas, surgical treatment, angiomatous meningioma*

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INTRODUCTION

The diagnosis of spinal angiomatous meningioma is extremely rare (representing about 1%) of meningiomas localized on the spinal cord, and the diagnosis is entirely histological [1]. Meningiomas can be found at various locations in the central nervous system and account for approximately 35% to 40% of benign tumors within it. Spinal meningiomas, according to literature data, account for 37.5% of all intradural extramedullary tumors in different regions of the spinal segments [2]. Spinal angiomatous meningiomas (SAMs) are rare and account for about 1-2% of all spinal cord meningiomas according to small clinical studies [1, 2, 3]. Angiomatous meningiomas are histologically characterized, as WHO grade I benign tumors, with abundant hyalinized blood vessels of variable size, an oval vesicular nucleus, and the presence of indistinct cytoplasm [4]. The proliferative index Ki67 in this type of tumors is low and is less than 1 (Ki67<1%), which once again confirms the benign nature of the tumor, from which it follows that total extirpation leads to good control of the pathological process [3, 4, 5, 6].

We present the case of a 44-year-old woman who gradually developed neurological symptoms on the part of conduction hypoesthesia, progressively worsening inferior paraparesis that reached paraplegia, while the patient underwent cranial surgical intervention for placement of a ventriculo-peritoneal shunt and subsequent clinical development of bacterial meningitis and secondary pneumonia of the lung. A magnetic resonance imaging (MRI) scan of the thoracic spine was performed a few weeks later and revealed evidence of chronic phase arachnoiditis. Eight months later, a new MRI of the thoracic spine was performed, which revealed cystic heterodense formations covering a wide thoracic region from the TH5-TH12 vertebral projections. The surgical treatment performed was aimed at histological verification of the process and subsequent medullary decompression.

CASE PRESENTATION

A 44-year-old woman presented with a history of headache, dizziness, and unsteady gait. On this occasion, hydrocephalus with normal mute was diagnosed and treatment included placement of a ventriculo-peritoneal shunt. The surgical intervention was performed in another medical institution and the data are based on the medical records submitted. The placement of the ventricular catheter was performed in a classical manner using the Frazier point and the distal part of the catheter was advanced into the epigastric region. A week later, complications occurred as the patient developed a severe septic condition, clinical and diagnostic data showed the presence of bacterial meningitis, with secondary superimposed pulmonary pneumonia. Due to the infection of the shunt system, as well as the inability to function properly, the latter was removed. A long course of antibiotic therapy and a stay in the intensive care unit followed. The patient recovered and was discharged upright and ambulating with a short distance walker. The neurological status was characterized by preserved tendon-suprascapular reflexes, no motor neurodeficit, muscle tone was represented by hypotonic muscle groups over the body, sensation was preserved, and gait was ataxic. The general somatic condition was stable and the patient did not require further treatment.

A few weeks later, the patient began to feel numbness and subsequently loss of sensation in her lower extremities. Within a week, her gait became more difficult, strength in the legs dramatically decreased, so she consulted a neurosurgeon, where the neurological status revealed: weakened tendon-supracondylar reflexes for the lower limbs, on examination for latent paresis-inferior flaccid paraparesis grade I to II, a conduction-type hypoesthesia distal to the umbilicus, with identical changes for both legs. MRI of the brain, cervical spine and sacral spine was performed, the reading showed adhesive processes in the thoracic lobe from a history of arachnoiditis and no subsequent surgical treatment was performed. The patient was discharged with no change in neurological status. Within a few weeks, the neurological status deteriorated, developing the clinic of inferior flaccid paraplegia with conduction-type hypoesthesia, to anesthesia distal to the rib arch (suppressed superficial but preserved deep sensation), pelvic urethral disturbances were of the incontinent type, necessitating placement of an indwelling urethral catheter, bowel motility was impaired, and manifestations of subileus developed as a result of neurogenic incomplete intestinal paresis. The patient was given laxatives to induce defecation. As a result of the sensory disturbances, trophic decubitus developed, the largest of which was noted in the sacral area. The ulcers were treated surgically over several months.

Eight months after the condition of paraplegia, the patient was admitted to the Department of Neurosurgery at the Pulmed University Hospital. The neurological status revealed that the patient had inferior spastic paraplegia, tendinous – suprascapular reflexes were alive with dilated reflexogenic zones, muscle tone was increased equally for both legs, pathological Babinski reflexes were registered bilat-

erally, superficial sensation was lost, deep sensation was partially preserved, the patient could not control the pelvic reservoirs, and the process of defecation. A new MRI of the thoracic spinal segment was performed, which revealed the presence of a heterodense formation in the T2 projections of the sagittal and axial planes, multiple cystic formations filled with liquoroequivalent fluid. In the T1 sequences, the image has a hypodense appearance, suppressed liquor signal, and T1 images with contrast material gave good information about homogeneous uptake of contrast material (Fig. 1, 2 and 3). Differentially, the diagnostic process was thought to be arachnoiditis with hydromyelia or meningioma located widely in the thoracic lobe.

In the course of diagnostic clarification, lung radiography (Fig. 4) and clinical and laboratory examinations were performed to assess the patient's general somatic condition.

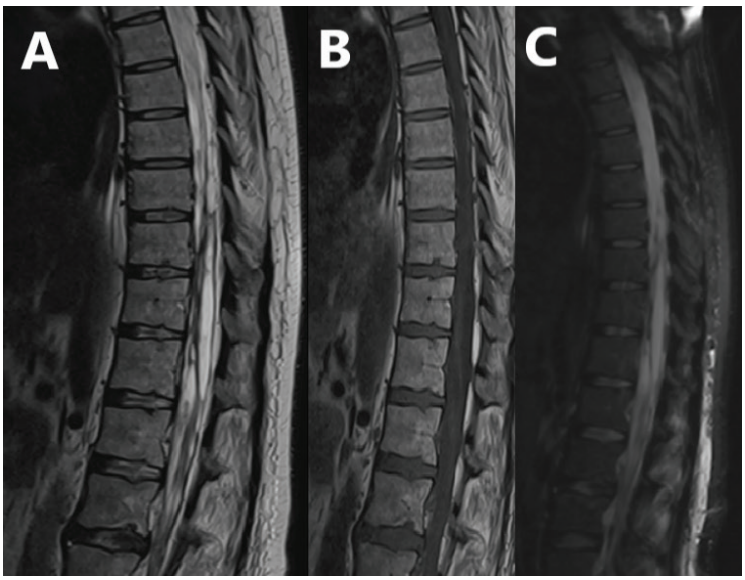

Fig. 1 (A, B and C). MRI of the thoracic spine. Panel A shows T2 sequence with heterogeneous characteristic of tumor process, presence of hyperintense liquorequivalent cysts. Panel B represents a sagittal section in T1 sequence. Panel C represents thoracic spine in tirm T1 sequence with signal suppression by adipose tissue

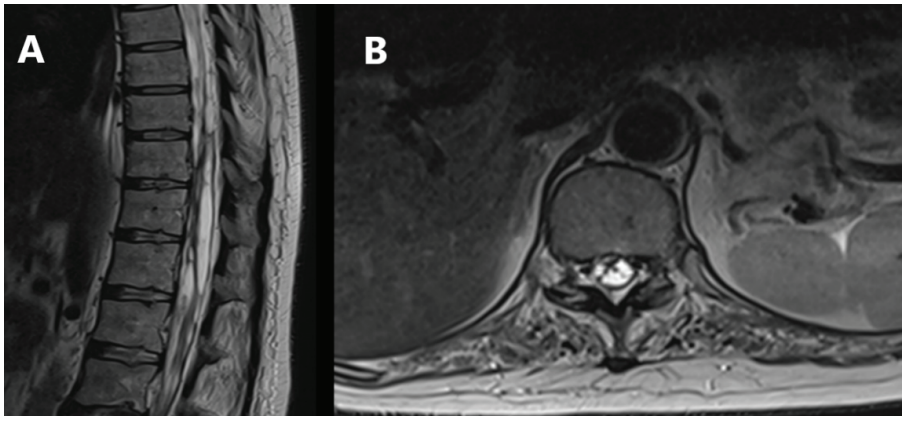

Fig. 2 (A and B). MRI of thoracic spine presenting sagittal and axial projections of new TH9-TH12 in T2 sequence

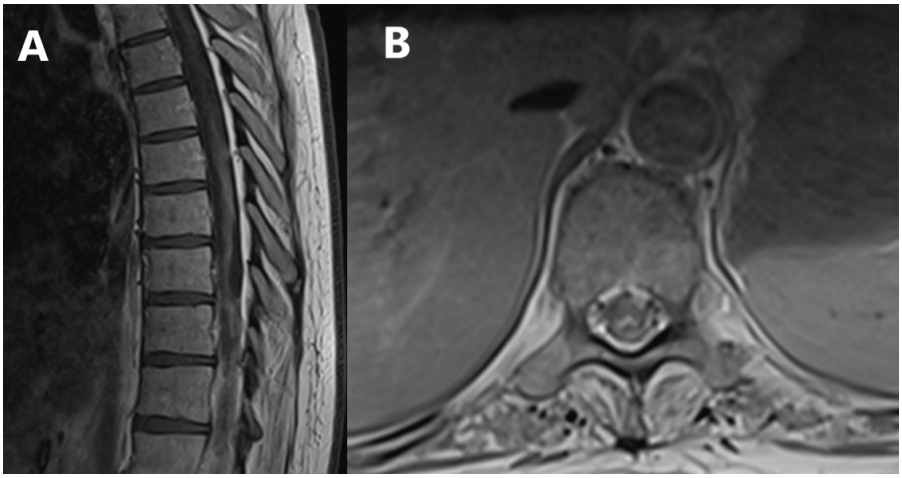

Fig. 3 (A and B). MRI of a thoracic spine in the sagittal and axial planes shows a T1 sequence with the application of contrast material (gadolinium) and the homogeneous uptake of the latter by the tumor specimen (hyperdense areas)

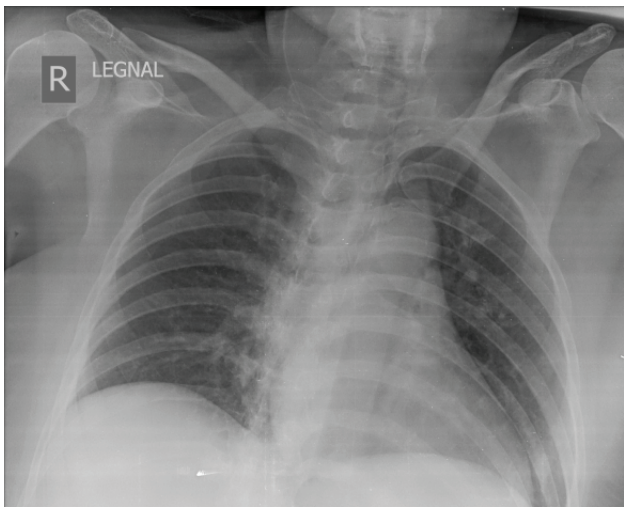

Fig. 4. Chest and lung radiography performed in the supine position with Kugel. The latter shows unenlarged hilar shadows, a thickened bilateral lung drawing. Preserved transparency of the parenchyma. No focal lesions and no evidence of active lung disease

We made a decision to perform surgical intervention – since at the levels of TH9-TH11 the tumor component was most pronounced to perform a total laminectomy of the TH9, TH10 and TH11 vertebrae, then to perform a median durotomy and extirpate part of the pathological finding, and if necessary to perform laminectomies of the upper vertebrae and to remove the process completely. Intraoperatively, an adherent formation was encountered for the dural surface. Macroscopically, the process was granular in texture, greyish-whitish in some places and raspberry-red in others, the firm texture. A clear border between the arachnoid and the underlying spinal cord was lost. The tumor formation was richly vascularized, and one of the cavities formed was fenestrated, and clear liquor was evacuated and sent for microbiological examination. There was no invasion of the liquor space by a bacterial causative agent. The presence of clear liquor indicates that there is a preserved subarachnoid space for liquor circulation to take place. In the area from the laminectomy performed, total tumor extirpation was achieved, but relative to the overall process encompassing the thoracic region, partial tumor extirpation was achieved (Simpson grade III). The medulla presented atrophic and given the neurological status, which persisted for several months, it was decided that it was not necessary to extend the extent of surgical intervention at this stage. Histology showed the presence of an angiomatous meningioma composed of capillary-type vascular lumena and hyalinization admixed with meningioma cells, few psammoma bodies and zones of hemorrhages (Fig. 5).

The patient remained in hospital for 16 days, during which the postoperative period was uneventful and the surgical wound healed primarily. Microbiological examination of urine was done where Escherichia coli was detected, antibiotic treatment was started as per the antibiogram with ciprofloxacin 100 mg./10 ml. at a dose of 2x400 mg. for a ten-day course. One month after the surgical intervention, the patient was in a wheelchair with impossible lower limb movements.

DISCUSSION

Angiomatous meningioma (AM) is a WHO grade I meningioma subtype, and spinal localization deter-

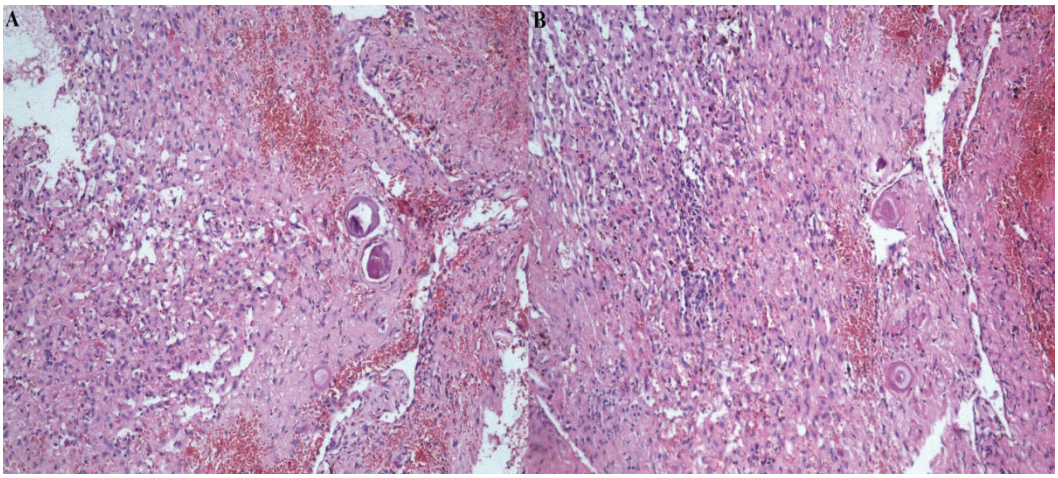

Fig. 5. The histological images demonstrate the typical features of an angiomatous meningioma, characterized by extensive vascular proliferation with numerous dilated capillaries and sinusoidal vessels lined by flattened endothelial cells. Tumor cells with eosinophilic cytoplasm and oval-to-spindle-shaped nuclei are arranged in a syncytial pattern around the vascular spaces, set within an edematous and occasionally hemorrhagic stroma. Concentric, eosinophilic structures resembling psammoma bodies are present, confirming their formation within the tumor. The findings highlight the significant vascular component and the presence of psammoma bodies, both characteristic of this rare meningioma variant (H&E staining, x100)

mines an incidence of approximately 1-2% of meningiomas located in the spinal canal [1]. Scientific publications in this field mainly include case reports or small series of clinical groups, which makes it difficult to systematize them by topographic-anatomical areas. The benign nature of the tumor also determines the good prognosis after their total surgical removal. The clinical course of thoracic meningiomas is characterized by a protracted course, and one of the first symptoms may be thoracic pain [2]. As the tumor volume increases to 64% of the spinal canal, symptoms on the medullary compression side are most common, with literature data indicating that motor neurological symptomatology is most prevalent in 92% of cases, followed by sensory dysfunction in 78% and gait disturbances in 42% of patients with thoracic spinal meningiomas. A significant proportion of cases (28%) developed bowel and bladder dysfunction when the tumor was grown [1, 2, 3]. The main symptoms are the result of mechanical compression of the spinal cord, disrupting the blood supply to the latter and leading to manifestations of myelopathy and irreversible neurological deficit with the manifestation of muscle spasticity and the presence of pathological reflexes, in the stage after the resolution of spinal shock [1, 4, 5, 6]. Risk factors for the occurrence of meningiomas in the spinal canal are defined as those of the surrounding environment- ionizing radiation, individual genetic factors. With the greatest reliability are the data in the presence of genetic mutations, such as neurofibromatosis type 1 and type 2 (NF1, NF2). The most common predisposition for the occurrence of multiple meningomatosis [10]. With the development of molecular genetics, it has been found that the genes affected in meningiomas are those in chromosome 22q12.2, which define NF type 2. Other genetic subtypes that can also lead to sporadic meningiomas are TNF receptor- associated factor 7, Kruppel-like factor 4 and others associated with various mutations on chromosome 22 [2, 4, 10]. In our case, the patient was 44 years of age and the risk factors that play a role are most likely related to hormonal expression on certain genes that activate the process of meningomatosis.

The diagnosis of spinal meningioma is made by means of imaging studies-computed axial tomography (CAT) and magnetic resonance imaging (MRI). The second method gives the most accurate information about the location of the tumor in the spinal canal, the size and the presence of edema of the medulla. In the noncontrast series on MRI, meningiomas appear isointense to the cerebral gray matter, and in the presence of calcifications to hyperintense. Placement of contrast material (gadolinium) results in homogeneous contrast enhancement of the tumor and well illustrates the feeding vessels, which allows planning of the surgical approach to the tumor mass. The two diagnostic imaging modalities (CTA and MRI) are complementary as they are non-invasive and provide different insights into the location and type of meningioma. CTA well illustrates the presence of calcifications in the tumor, such as hyperdense areas, thereby determining its density. MRI provides evidence of the relationship of the tumor to the dural surface and its blood supply. Extradural forms of meningiomas are also reported in the literature in rare cases. The differential diagnosis of these tumors, covers a wide range of metastatic processes, with a rare case for metastasis to the vertebral bodies being a thoracic melanoma that results in medullary compression on extension to the spinal canal. The presence of a low density of the tumor mass determines the possibility of using an ultrasound aspirator during tumor excision, which dramatically reduces the iatrogenic impact on the spinal cord [2, 3, 10, 11, 12, 15, 16, 17].

In meningiomas of the spinal canal, the histological diagnosis plays a major role, which determines the type of tumor and its degree of malignancy. In our case, there was more than 50% presence of blood vessels in the histological material, as well as the presence of psammoma corpuscles, which warranted the diagnosis of angiomatous meningioma (AM). In these tumors, morphology showed the presence of marked hyalinization, scattered blood vessels of different caliber, and the presence of meningothelial cells with round to oval nuclei and eosinophilic cytoplasm. Most often the cells are arranged as nests. The proliferative index Ki67 in WHO type I tumors is low, which determines a low rate of tumor recurrence with en bloc resection and achievement of Simpson grade I [11, 12, 13, 16].

In the differential diagnosis of meningiomas of the spinal canal, as well as in our clinical case, the presence of spinal adhesive arachnoiditis enters into consideration, which can be excluded by histological examination. The clinical manifestations of arachnoiditis resemble those of angiomatous meningioma, as do the MRI findings, as in our case. The morphology of arachnoiditis is associated with the presence of fibrous cells, hyalinization of the arachnoid and absence of abundant blood vessels and the presence of meningothelial clumps. These distinguishing features are sufficient to make the diagnosis of AM instead of spinal arachnoiditis [9].

Surgical treatment for symptomatic meningiomas includes devascularization of the feeding vessels, microsurgical dissection from the adjacent spinal cord and en bloc removal of the formation, com-

bined with neuronavigation and neuromonitoring, resulting in minimal damage to the intact spinal cord [10]. In our case, total removal of the tumor was not possible due to the location of a broad base in the thoracic compartment and the presence of long-standing persistent debilitating neurological symptomatology. When total removal of the meningioma was not possible and Simpson's resection was classified in a higher grade, radiosurgery came into consideration [9, 10, 11]. Functional activity of patients with thoracic meningiomas is determined by the Karnofsky quality of life scale, as well as the modified McCornick and Frankel scales, which define sensory and motor deficits before and after surgery. The classic way is to follow up patients with thoracic meningiomas initially at 3 months and then at 6 months by imaging and clinical staging [11, 12].

CONCLUSIONS

Angiomatous meningiomas are rare, benign tumors that can cause a variety of neurological symptoms due to compression of surrounding structures. Spinal localization of this type of tumors is extremely rare and of clinical and histological interest. Early diagnosis by MRI and appropriate treatment are essential to improve patient's quality of life. Given the benign histological variant of this type of meningiomas, surgical treatment consisting of total extirpation of the tumor with subsequent clinical follow-up are sufficient to achieve good tumor control. The present case is illustrative in that the ataxia most likely occurred as a result of a change in lower limb sensation from initial medullary compression and was misinterpreted as a symptom of the hydrocephalus syndrome. Meningiomas are slow-growing tumors and in most cases a definitive diagnosis requires several months from the initial symptoms.

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Consent for publication: *Consent form for publication was signed by the patient/relatives and collected.*

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