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CASE REPORT

A RARE NEUROSURGICAL CHALLENGE: BRAIN MENINGIOMA WITH EPIDURAL HEMATOMA

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Abstract. *Brain meningiomas are predominantly benign extracranial brain tumors arising from the arachnoid cells of the meninges. According to their anatomical localization and trapping site, they are arranged in different groups. The World Health Organization categorizes meningiomas according to their malignancy and classifies them into three types, each of which includes a different histological type of meningioma. The vast majority of meningiomas are benign and recurrence depends largely on surgical resection and subsequent radiotherapy. The main methods in the diagnosis of central nervous system meningiomas are computed axial tomography and magnetic resonance imaging enhanced with contrast material, the radiological hallmarks of meningiomas being the presence of homogeneous filling of the tumor with contrast and a „dural tail", which are among the key pathognomonic features of an extraaxial tumor. Surgical treatment is rarely challenging, except in cases of a richly vascularized tumor, extension of the latter to venous sinuses, and atypical localization of meningiomas. Achieving a Simpson grade I or II resection significantly reduces the risk of tumor recurrence. A rare type of complication is postoperative epidural hematoma at the site of the performed craniotomy or at a different site from it, which leads to a rapid and progressive deterioration of the general brain condition. We present a case of a 46-year-old woman with invasive breast cancer and meningothelial meningioma located frontally (parasagittally) on the left with marked compression of the underlying brain parenchyma, leading to general cerebral and focal neurological symptomatology. Simpson grade I removal of the meningioma was performed. Postoperatively, the patient developed a state of disseminated intravascular coagulopathy, with worsening of the general cerebral condition to coma, from the development of a left parietotemporal epidural hematoma. Timely diagnosis and operative treatment led to a favorable outcome from the complications that developed.*

Key words: *meningioma of the brain, histology of meningiomas, epidural hematoma, surgical complications, coagulation disorders and epidural hematoma, benign brain tumors, disseminated intravascular coagulopathy*

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INTRODUCTION

Meningiomas are the most common group of intracranial brain tumors, accounting for over 1/3 of all adult-onset primary central nervous system tumors [1, 2]. Over 80% of them are benign (WHO grade I), showing a predilection for women (3:1) and advanced age [3, 4]. Originating from the arachnoid cap cells of the arachnoid mater, these neoplasms show significant differences according to their anatomic location, from asymptomatic course to marked neurological deficit [4]. The gold standard for their diagnosis is magnetic resonance imaging (MRI) and computed tomography (CT) with contrast, and they are sometimes discovered incidentally during neuroimaging studies on another occasion [5]. Surgical resection of this type of tumor remains the main method of treatment, with greater radicality corresponding to a reduced risk of recurrence [5, 6]. One of the rare postoperative complications (about 1%) is the occurrence of epidural hematoma, associated with the young age of the patients and rapid drainage of a large amount of cerebrospinal fluid (CSF) [7, 8]. Such a potentially fatal complication would mar even the most precisely performed meningioma resection. We report a case of a patient with a large meningioma located frontally on the left with a rare surgical complication – epidural hematoma and progressive worsening of general neurologic functions and vital signs.

CASE PRESENTATION

A 46-year-old woman presented with complaints that had debuted several months ago – progressively increasing headache and dizziness. Gradually she became confused, disoriented, her speech became difficult, she had difficulty pronouncing words, her gait was unsteady, and she began dropping objects, while feeling her right limbs weaker and clumsier. Tremor and hypesthesia appeared in her right limbs. On this occasion the woman visited a neurosurgeon and on examination, she was alert but confused and disoriented to time and place. Clinical evidence of a general cerebral hypertensive syndrome, combined with a frontal apathetic-abulic syndrome was found. There was no evidence of meningo-radicular irritation, cranial nerves were without abnormalities. Latent paresis samples revealed latent right-sided hemiparesis combined with hemihypesthesia. The tendinous and supraosseous reflexes were pinched for right limbs, pathological Babinski's symptom was found on the right. Muscle tone was moderately increased for the right limbs. Superior cortical functions presented with partial motor aphasia.

The comorbidity that the patient reported of was histologically proven invasive moderately differentiated ductal carcinoma for which she was undergoing chemotherapy. Immunohistochemistry from the latter showed the following markers ER- 75% chance of response to therapy, PR- 75% chance of response to therapy, Her2- /1+/- negative, Ki67 < 10% positivity, E- cadherin- positive. The medical history was also significant for left ovarian cyst, right sided severe ureterohydronephrosis and uterine fibroid.

Magnetic resonance imaging (MRI) of the brain showed evidence of a hyperdense T2 sequence parasagittal frontal left tumor formation, with the characteristics of a meningioma. The latter, according to its localization, has MRI characteristics of a mixed meningioma with a focal site along the convex dura and falx cerebri. Its approximate dimensions measured in axillary sections of T2 sequence without placement of intravenous contrast material were 6.83/5.02 cm, and in sagittal sections of T1 sequence, where the meningioma has a hypodense characteristic in relation to the brain parenchyma, the dimensions measured from the frontal to the coronal suture and from the calvaria to the skull base were 7.37/5.68 cm (Figure 1).

To confirm the diagnosis of meningioma of the brain, MRI examination was also performed with placement of gadolinium contrast material (Figure 2).

Laboratory tests were performed and showed no abnormalities, the coagulation status of the patient on admission to the neurosurgery department showed: prothrombin time (sec) – 13.9, prothrombin time (%) – 90.0, international normalized ratio (INR) – 1.08. Biochemical tests were without deviation from the norm, consultative examination by a cardiologist showed no contraindications for surgical treatment. Electrocardiogram revealed sinus rhythm without repolarization changes. A standard preoperative procedure in our department is to perform pulmonography in patients undergoing general intubation anesthesia. The latter revealed a symmetrical chest, unenlarged hilar shadows, preserved transparency of the lung parenchyma, diaphragms were normally configured, and the cardiac shadow was unenlarged. The report found no radiological evidence of active lung disease.

After discussion by the lecturing team, it was decided that this type of tumors is strongly indicated for surgical treatment. The patient was placed supine on the operating table under general intubation anaesthesia, the head was fixed on a three-point Mayfield and rotated to the right. A stentart frontal craniotomy was performed on the left, parasagittally, the dural meninges infiltrated by the tumor were encountered,

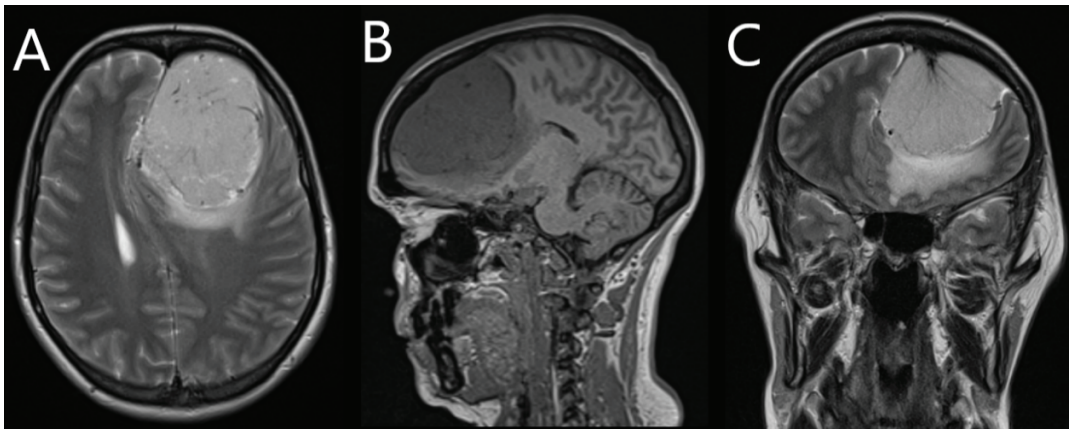

Fig. 1 (A, B and C). Native brain MRI. Panel A shows an axial slice in T2 sequence from MRI of the brain showing hyperdense formation in the left frontal with dural tail and characteristic meningioma, perifocal cerebral edema and dislocation of midbrain structures. Panel C shows a T1 sequence of a frontally located hypodense formation in sagittal section. Panel C shows a coronal hyperdense image in T2 sequence through the meningioma shad

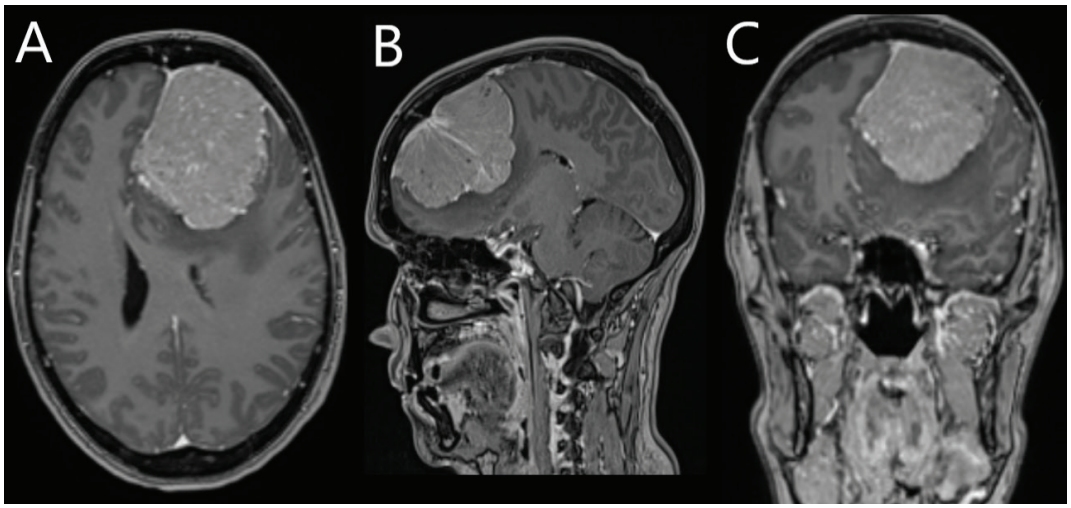

Fig. 2 (A, B and C). MRI of the brain in T1 sequence of axial (panel A), sagittal (panel B), and coronal (panel C) sections with contrast enhancement (gadolinium) are shown. A hyperdense formation is seen, with multiple blood vessels and homogeneous filling of the latter, dural tail and imaging features of a meningioma

the latter was transected and median enucleation of the tumor was proceeded with the aim of reducing its growth under microscopic control. Macroscopically, the appearance of the tumor was grayish-white in colour, with a granular structure, aspirated in some places and non-aspirated in others with the presence of calcifications, encapsulated and with a clear border in the subarachnoid plan. Under microneurosurgical control, the tumor was devascularized from its feeding vessels, detached from its capture site along the falx, the convecting dura that had been infiltrated by the tumor was excised, and thus Simpson grade I removal of the meningioma was performed. Dura plasty with synthetic meninges was performed. The patient was transferred to the intensive care unit, where she was extubated an hour later. Neurologically, she was lucid, satisfactorily adequate, with no worsening of baseline neurological status. Laboratory investigations such as complete blood count showed some abnormalities thrombocytosis – 989.109 (reference values 100.109-400.109), biochemical investigations were unchanged, coagulation status showed significant postoperative changes – prothrombin time (sec) – 26.4, prothrombin time (%) – 38.0, international normalized ratio (INR) – 2.14; fibrinogen – 1.55 mmol/l; D-dimer – 1.11 ng/ml (reference values – 100-200 ng/ml). Laboratory parameters showed the development of disseminated intravascular coagulopathy (DIC-syndrome), fulfilling the criteria of the ISTH scale for the development of DIC-syndrome. Our patient had a total score equal to 5, confirming the development of DIC-syndrome. Two hours after the surgical intervention, the patient became unconscious, comatose, bradycardic with a sudden

collapse of hemodynamics, necessitating repeated intubation and administration of noradrenaline due to hypotension and inability to control her vitals. A control CTA of the brain was performed, which revealed an epidural hemorrhage parietotemporally on the left (Figure 3).

Emergency surgical treatment was performed by forming a new bone flap temporoparietally on the left and encountering an epidural collection represented by hemocoagulomas about 4 cm thick and 7/7 cm in area. The haemocoagulums were removed and multiple duroperiosteal sutures were placed in the setting of epidural bleeding, thereby controlling bleeding in the operative field. A subdural revision was performed intraoperatively where no haemorrhagic collection was found. The most likely cause of this rare complication was the patient's development of DIC syndrome, coupled with a rapid decrease in intracranial pressure after removal of the tumor mass.

Postoperatively, the patient was extubated after 24 hours, regained consciousness, and no further neurological complications were found during the physical examination after extubation. A contralateral control CTA of the brain was performed (Figure 4).

After a thorough examination the histological result confirmed the diagnosis of meningothelial meningioma (Figure 5). Meningothelial meningioma is the most common histological subtype of meningioma. It is mostly presented by sheets of uniform, oval cells with indistinct cell membranes, giving an architecture of whorls. The nuclei are round to oval with finely dispersed chromatin.

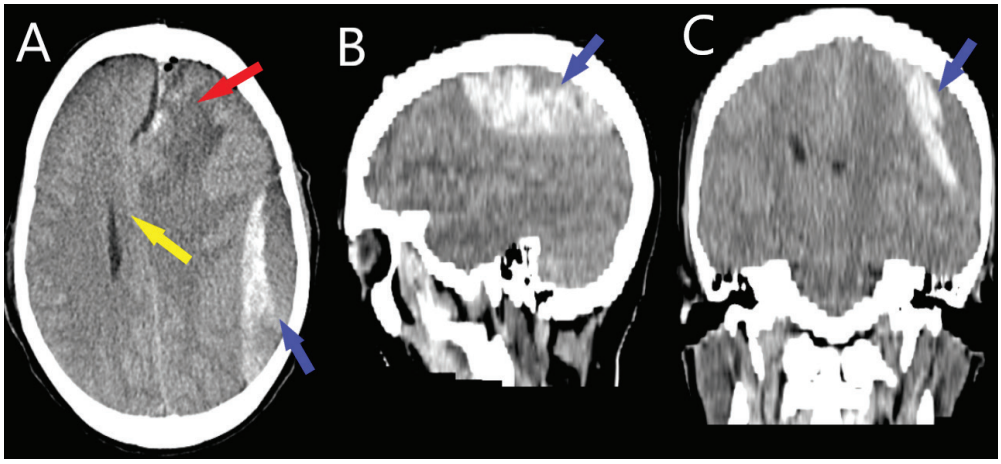

Fig. 3 (A, B and C). CT scan of the brain presenting three projections (axial, sagittal and coronal) plane with the presence of a hyperintense collection in the left parieto temporal with evidence of epidural hematoma. Panel A, B and C with blue arrow shows the epidural hematoma. The red arrow in Panel A shows postoperative edema at the site of the extirpated meningioma, the yellow arrow in Panel A, shows the dislocation of midbrain structures, which is 0.7 mm from the midline

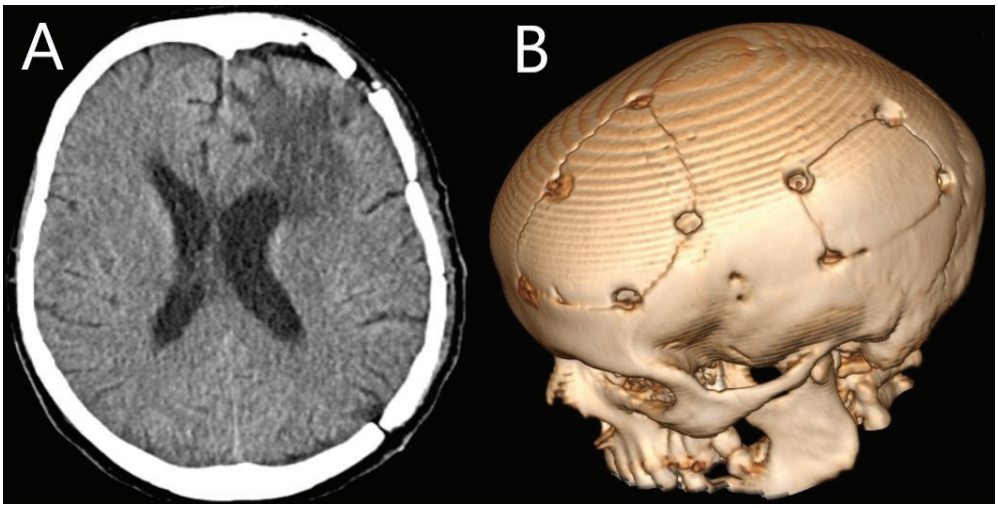

Fig. 4 (A and B). CTA of the brain postoperative after meningioma extirpation and epidural hematoma evacuation. Panel A shows hypodense areas anteriorly from persistent perifocal edema at the site of the extirpated tumor. Panel B shows a 3D reconstruction of the skull and craniotomy sites

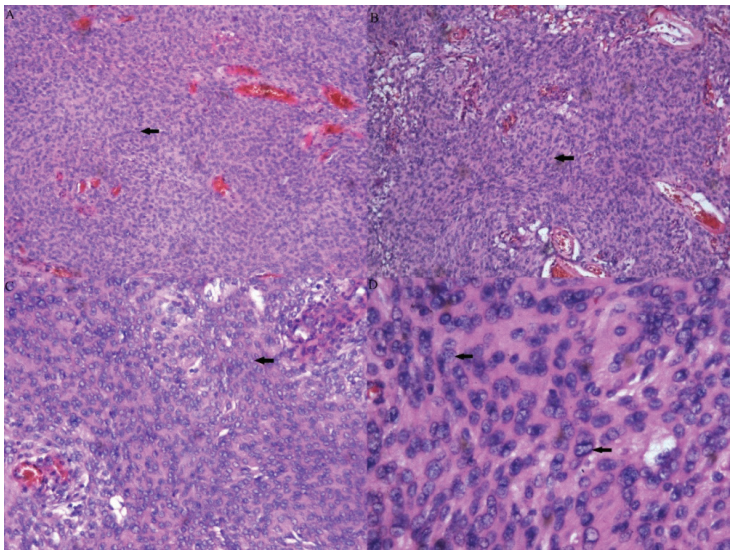

Fig. 5. A. Histological view of the specimen, H.E. staining, original magnification x 100; **B.** Whorls of meningothelial cells, H.E. staining, original magnification x 100; **C.** Lobulated architecture of meningioma, H.E. staining, original magnification x 200, **D.** Round to oval nuclei with finely dispersed chromatin, H.E. staining, original magnification x 400

Post-surgery, the patient spent fifteen days in the neurosurgery ward where she had daily physiotherapy, which enabled prompt verticalization and helped the patient move independently. She was discharged in a clear conscience, contact, transiently confused and with improved motor neurodeficit. The patient was followed up by a neurosurgeon annually via brain MRI. MRI at the second year after tumor extirpation showed no evidence of recurrence (Figure 6).

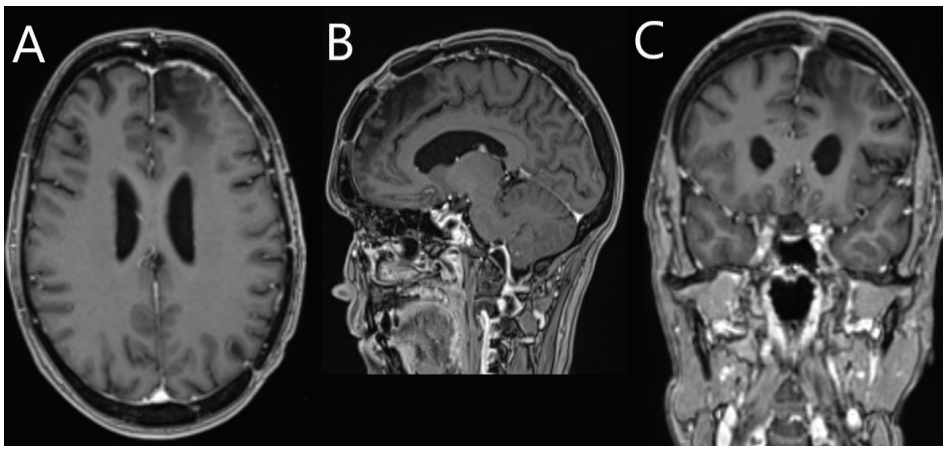

Fig. 6 (A, B and C). MRI of the brain with contrast (gadolinium). Shows axial, sagittal, and cortical sections of the cerebellar mosaic in T1 sequence showing no meningioma recurrence

DISCUSSION

Meningiomas of the brain are the most common benign tumors of the central nervous system (CNS). Studies of large cohorts of people in the US have shown that they account for about 37.6% of all primary brain tumors and 53.2% of all benign CNS tumors [3, 4]. Originating from the arachnoid cap cells of the leptomeninges, they often arise near the dural venous sinuses-sites of clusters of pacchionian granulations, rich in arachnoid cells. The location of meningiomas largely determines the appearance of focal neurological symptoms. The most common site of meningiomas is the lateral convexity of the hemispheres, parasagittal, sphenoidal, and middle cranial fossa, frontobasal, posterior cranial fossa [3]. The classification of the World Health Organization (WHO) divides meningiomas into 3 degrees of malignancy according to the histopathology of the tumor or its subtype, with the meningothelial variant being the most common and falling into grade I, characterized by slow growth and the presence of meningothelial concentric formations, syncytial cells with rounded nuclei, eosinophilic cytoplasm, and pseudoinclusions [3, 9, 10]. AKT1, TRAF7 and SMO gene mutations are frequently expressed in this histological type of meningiomas [5]. Exposure to ionizing radiation is one of the key factors that significantly increases the risk of meningioma. The incidence of these tumors is also associated with female sex hormones, as well as breast carcinoma, but the exact relationship and etiology remain uncertain [1, 11]. Genetically, meningiomas are a common manifestation of neurofibromatosis type 2 (NF2), inherited in an autosomal dominant manner, as well as some other rarer hereditary syndromes [1].

The diagnosis of meningiomas is based on imaging studies (MRI and CT) performed in case of the appearance of neurological symptoms. As with other brain tumors, it mainly depends on the location of the tumor mass, the spread to the adjacent structures and the rate of growth. About 1% of meningiomas can be found incidentally, without pronounced symptoms, during another imaging study, and most often they are indicated for surgical treatment [5]. Meningiomas on CT images appear as circular, hyperdense, extra – axial structures that have grown into the dura mater, clearly demarcated from the surrounding brain parenchyma. Due to their slow growth, the accumulation of calcium deposits is often observed. CT is the best method for detecting infiltration of the cranial bones adjacent to the meningioma. Their typical magnetic resonance image is hypo- or isointense on T1 sequence and iso- or hyperintense on T2. The presence of contrast material in both imaging methods significantly improves tumor margins and helps differentiate intratumoral formations such as cysts, hemorrhages, and cellular breakdown [12]. The observed dural tail in our clinical case occurs in nearly 72% of meningiomas, helping to distinguish them on imaging studies from other tumors. Although often associated with meningiomas, it should not be taken as a definitive pathognomonic sign [12].

The main method of treating meningiomas remains surgical resection, which can be supplemented by radiotherapy and chemotherapy. For asymptomatic, small tumors (≤ 3 cm), the approach may be wait – and – see, with follow – up MRIs once a year [1]. Due to the significantly larger size of the tumor in our clinical case, as well as the presence of focal and general neurological symptoms, a total resection of the meningioma was performed, as well as of the convexity dura infiltrated by it, corresponding to grade I on the Simpson scale. Simpson II and III tumor resections are less favorable and may increase the risk of recurrence, with Simpson IV having been shown to be the most unfavorable long-term tumor resection associated with a significantly increased recurrence rate [1, 6]. In Simpson V, decompression with biopsy is performed, although it is questionable how objective the distinction between grade IV and V is [13]. One of the potential complications of any craniotomy is the development of postoperative epidural hematoma (PEH), seen in about 1% of cases. It can develop immediately below the site of the operative access, which is most common in the development of PEH, or at some distance, as in our case (left frontal craniotomy vs left parietotemporal PEH) [7]. While the pathophysiology of PEH is not fully understood, it is assumed that the abundant evacuation of CSF, the elevation of the patient's head above 30 degrees on the operating table and the displacement of the brain parenchyma caudally, under the influence of gravity, and the sharp drop in intracranial pressure, are the factors causing detachment of the firmly attached dura mater to the overlying bone, with subsequent traction of the meningeal vessels and bleeding [8, 14]. An associated risk factor is the relatively young age of our patient – 46 years old. The vast majority of the described cases of PEH in the literature are in young patients, the probable reason for this being the weaker adhesion between the dura and the bone, as well as the increased pachymeningeal elasticity [7, 14]. The immediate development of disseminated intravascular coagulopathy (DIC syndrome) after the end of the surgical intervention is the other important moment in the pathogenesis of the epidural hematoma in our case. A probable cause of this is the release of thromboplastin, abundant in the brain parenchyma, as well as procoagulant tissue elements into the circulating blood. These coagulation factors are frequently expressed in various solid tumors, including meningiomas, as Eom et al. reported their high association with breast adenocarcinoma, as well as the initiation of DIC syndrome following surgical intervention [15]. All these factors, including those described above – young age and removal of a large tumor mass, are the probable cause of the development of PEH in our patient. As a prevention of this potential complication, it can be recommended to avoid copious and rapid evacuation of CSF, readiness for transfusion of fresh frozen plasma and platelet mass in case of DIC syndrome, as well as the early detection of PEH in the postoperative period by making control CTs, and immediate evacuation in case of the latter [7, 15].

CONCLUSIONS

Meningiomas are the most common benign brain tumors, and their clinical appearance can occur after a long latent period of growing over many years without causing any symptoms or can be an incidental finding, when performing an imaging study on another occasion. Although the removal of the majority of them does not represent a problem from a surgical point of view, the postoperative epidural hematoma accompanied by DIC syndrome – a rapidly progressive and potentially fatal complication – must be taken into account. The case described by us is rarely seen in the literature and presents the clinical course and prerequisites for this possible complication. Slow and limited drainage of CSF, obtaining postoperative imaging studies, and the readiness of the surgical

team for rapid revision determine the favorable outcome in such a clinical scenario.

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Ethical statement: *This study has been performed in accordance with the ethical standards as laid down in the Declaration of Helsinki.*

Informed Consent for a Clinical Case: *Written informed consent was obtained from the patient for the publication of this case report, including any accompanying images.*

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