

Bilateral chronic subdural haematomas in a patient with meningioma of the superior sagittal sinus – case report and pathophysiological study

Oponiak zatoki strzałkowej górnej z towarzyszącymi obustronnymi przewlekłymi krwawkami podtwardówkowymi – opis przypadku i studium patofizjologiczne

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Abstract

Bilateral chronic subdural haemorrhage accompanying meningioma is a very rare clinical condition. We present a case of a 69-year-old female patient with large meningioma completely obliterating the posterior third part of the superior sagittal sinus with accompanying bilateral chronic subdural haematomas. Three anatomical zones of venous collateral circulation were revealed by the preoperative digital subtraction angiography. The tumour and haematomas were removed completely with no major complications.

The most likely pathomechanism of the development of bilateral chronic subdural haematomas was venous hypertension caused by an occlusion of major cerebral venous trunks. As a result of a minor thrombotic incident or insignificant head injury, the distended veins of collateral circulation that were volumetrically burdened could have been damaged. Patients with large tumours occluding the superior sagittal sinus, who did not qualify for or refused surgery, should be carefully monitored clinically and neuroradiologically because of possibly increased risk of an intracranial haemorrhage.

Key words: superior sagittal sinus meningioma, subdural haematoma, cerebral collateral circulation, sagittal sinus occlusion.

Streszczenie

Przewlekłe krwawienie podtwardówkowe towarzyszące oponiakowi wewnątrzczaszkowemu jest stwierdzane niezwykle rzadko. W pracy zaprezentowano przypadek 69-letniej pacjentki, bez istotnej przeszłości chorobowej, u której stwierdzono olbrzymi oponiak sklepiści czaszki całkowicie zarastający światło tylnej jednej trzeciej zatoki strzałkowej górnej współistniejący z obustronnymi mnogimi przewlekłymi krwawkami podtwardówkowymi. Przedoperacyjna cyfrowa angiografia subtrakcyjna wykazała obecność trzech stref krążenia obocznego kompensującego niedrożność zatoki strzałkowej górnej. Guz oraz przewlekłe krwawiki podtwardówkowe usunięto doszczętnie, przebieg pooperacyjny był pomyślny.

Jedną z opisywanych przyczyn powstawania przewlekłych krwawików podtwardówkowych w przypadkach powolnego zamknięcia dużych żył mózgowych jest pęknięcie przeciążonych objętościowo naczyń krążenia obocznego. Opisywany przypadek to prawdopodobnie pierwszy dobrze udokumentowany przykład potwierdzający słuszność tej tezy. Pacjenci z guzami zamykającymi światło zatoki strzałkowej górnej, którzy nie są poddawani leczeniu operacyjnemu, powinni być monitorowani klinicznie i radiologicznie ze względu na zwiększone ryzyko krwawienia wewnątrzczaszkowego.

Słowa kluczowe: oponiak zatoki strzałkowej górnej, krwawk podtwardówkowy, mózgowie krążenie oboczne, niedrożność zatoki strzałkowej.

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Introduction

Intracranial haemorrhage affects 4-10% of patients with brain tumours, most frequently those with glioblastoma multiforme and intracranial metastases. It has been established that intracranial bleeding accompanying meningioma is very rare [1-3]. An exceptionally unique finding is subdural haemorrhage. In 2003, Bruno *et al.* [1] presented data on 141 cases of meningiomas accompanied by intracranial haemorrhages and revealed 24 cases of subdural haemorrhages. In only three cases were haematomas bilateral.

One of the many pathomechanisms of this type of bleeding, proposed by Kohli and Crouch, is venous hypertension in the collateral circulation induced by an occlusion of large venous vessels [4]. Nevertheless, the available literature does not contain any particular reports confirming this theory.

In this paper we describe a case of bilateral chronic subdural haematomas accompanying a meningioma of the posterior third part of the superior sagittal sinus (SSS). Particular attention has been paid to the pathogenesis of the haematoma development.

Case report

A 69-year-old female patient was admitted to the Department of Neurosurgery of Wrocław Medical University due to headaches lasting more than three weeks. The patient was hospitalized four years earlier due to persistent lower extremity numbness lasting more than one year. Magnetic resonance imaging (MRI) scans of the head revealed a tumour of SSS with no features of intracranial haemorrhage. The patient refused surgery. Lower extremity numbness regressed spontaneously and the patient refused further medical treatment or diagnostic tests. Computed tomography (CT) performed on admission to the department revealed a tumour in the SSS comparable in size to the one observed in the examination performed four years earlier. Additionally, bilateral chronic subdural haematomas were discovered.

Examination

Physical examination did not reveal any significant abnormalities, except bilateral discolouration of the eyelids, most likely caused by dilatation of small veins. Laboratory tests revealed normal values of the ionogram, haematological and coagulation parameters. There was no sign of either a major or a minor trauma in the anamnesis.

The MRI of the head revealed a poorly vascularized tumour in the posterior third of the SSS of a grape-like shape, infiltrating the dura mater and the bones of the cranial vault around the SSS. The other radiological findings were bilateral chronic subdural haematomas of different densities in the frontal and parietotemporal regions, which did not have anatomical communication with the tumour (Fig. 1). The MRI did not show any direct links between the tumour and any of the chronic subdural haematomas. Digital subtraction angiography (DSA) of the cerebral arteries with the end-to-late venous phase was also performed. It revealed a complete occlusion of the posterior third of the superior sagittal sinus at the length of 11 cm with the development of a venous collateral circulation. Three anatomical zones of the venous collateral circulation were found: 1) veins draining into the anterior third of the SSS which formed a connection with the superior ophthalmic vein, with the angular vein and temporal veins draining into the facial vein; 2) veins draining into the parietal lacunae of the SSS, connected with veins of the sphenoidal group draining into the cavernous sinus; 3) veins draining primarily into the posterior SSS connected with veins of the tentorial group draining into the transverse sinuses (Fig. 2).

Operation

The patient was operated on in a semi-sitting position. Craniotomy allowing the exposure of the tumour and posterior parts of the two largest subdural haematomas over both cerebral hemispheres was performed. A tumour infiltrating the dura mater and the SSS in its posterior third at a length of roughly 10 cm was exposed. The tumour did not reach the venous tributaries. It caused a complete occlusion of the SSS in the affected segment.

The tumour was excised completely and all significantly dilated venous vessels of the cerebral cortex in the tumour vicinity were spared. Because there was efficient venous collateral circulation, the SSS was not reconstructed [5,6]. The proximal and distal stumps were secured with sutures. Encapsulated chronic subdural haematomas were opened bilaterally in the temporal regions. The presence of numerous compartments with different degrees of haematoma evolution was discovered.

Pathological findings

The routine histopathological study revealed a highly pleomorphic tumour with numerous microcysts, psammoma bodies and a large fibrous component

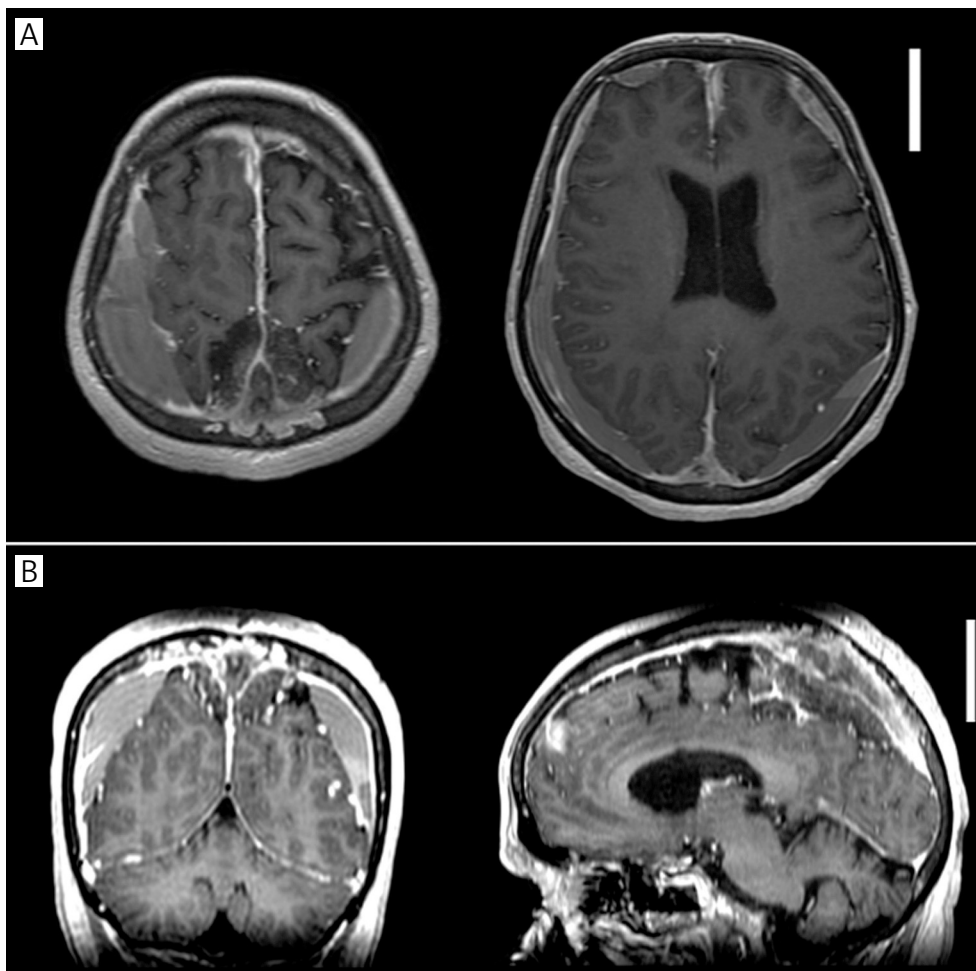


Fig. 1. A) Axial T1-weighted gadolinium-enhanced MRI. B) Coronal and sagittal multiplanar reconstructions. The bar indicates 4 cm

(Fig. 3A). The neoplastic cells were strongly positive for vimentin and the epithelial membrane antigen (EMA). Tumour cells showed a minor immunoreactivity for CD34 (Fig. 3B).

No tumour infiltration was observed in the obtained fragments of the dura mater.

Postoperative course

The patient tolerated the operation very well and was discharged 7 days after the procedure. A follow-up neurological examination performed before discharge and three months after discharge revealed no neurological deficits. The MRI performed three months after the surgery revealed no signs of subdural haemorrhage or tumour regrowth.

Discussion

According to Rocco *et al.* [3], the incidence of intracranial bleeding accompanying meningiomas in a large series of patients ranges from 0 to 2%. Subdural haematomas are particularly rare [1].

Currently it is known that the most common source of bleeding is the tumour itself or the cerebral venous vessels that have been infiltrated by the tumour [2]. In our case, despite the heterogeneous histological structure of the tumour, no neoangiogenesis intensification was observed. In addition, preoperative and intraoperative examinations did not reveal the presence of a well-developed tumour vasculature. Therefore microhaemorrhage from the tumour seems to be very unlikely in this particular case. Such a premise is additionally

strengthened by the finding that all haematomas were formed at a distance from the tumour without a direct connection with it.

Hypertension in the venous collateral circulation may also be the reason for the development of intracerebral haematomas, especially in the direct vicinity of the tumour [7,8]. The formation of the collateral circulation in the case of a tumour occluding the lumen of the SSS is a common condition [9]. In our case, end-to-end connections between the veins of the superior sagittal group and the veins of the tentorial and sphenoidal groups were formed [10] as well as with the superior ophthalmic vein and the facial vein (discoloration of the upper eyelids). This finding has not been described previously. Slow tumour growth during a period of 4 years enabled the formation of the collateral circulation. In 1991, based on the observation of 37 SSS meningioma cases, Czernicki *et al.* [11] did not find any correlation between an occlusion of the sinus and the presence of brain oedema or features of increased intracranial pressure. The authors concluded that slow tumour growth allowed the efficient collateral circulation to develop. However, they admit that dilatation and distention of veins of the collateral circulation were usually found intraoperatively. Intraoperative observations made by Czernicki *et al.* [11] and results of experimental studies by Ueda *et al.* [12] indicate that the occlusion of the SSS leads to increased pressure in the system of the superficial and bridging veins. A limited number of venous vessels of the collateral circulation have to receive the volume of blood that normally passes through the superior sagittal sinus – approximately 400 ml/min [13]. In the described case, damage of overloaded veins could have occurred in a similar manner as in the cases of a rupture of arterialized bridging veins of arteriovenous malformations [14,15].

The large number and bilateral location of haematomas in the areas where the collateral veins were particularly well developed and visible in DSA (the parietal and frontal areas) and the lack of other documented reasons for a haemorrhage can support this thesis. As a result of a minor thrombotic incident or insignificant head injury, the distended veins that were volumetrically burdened could have ruptured and there might have been long-term, mild bleeding. This can be confirmed by the fact that the haematomas were encapsulated and divided into smaller compartments of different resolution stages, which was observed radiologically and intraoperatively.

This case study supports the theory of venous hypertension which is caused by neoplastic occlusion of ma-

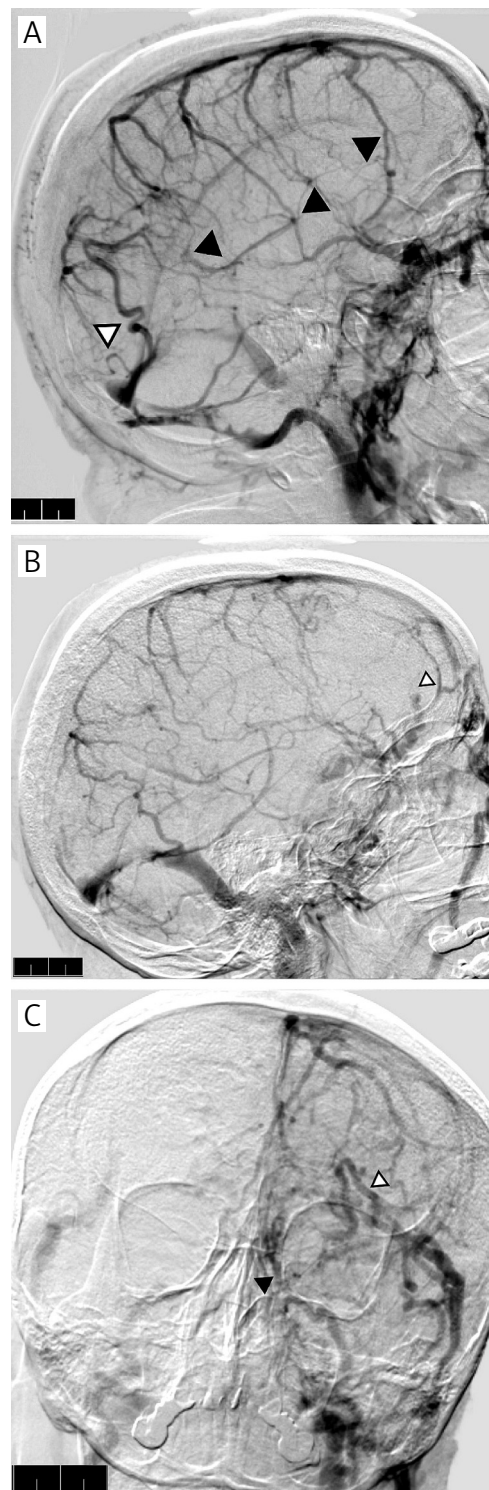


Fig. 2. Digital subtraction angiography of cerebral arteries, late venous phase. A) Lateral projection; no blood flow in the superior sagittal sinus at the tumour site. Selected end-to-end anastomoses are marked with arrows. B) Lateral projection: the arrow indicates connections between the veins of the anterior part of the superior sagittal sinus and the superior ophthalmic vein. C) AP projection; anastomoses of superior ophthalmic vein. The bar: 2 cm

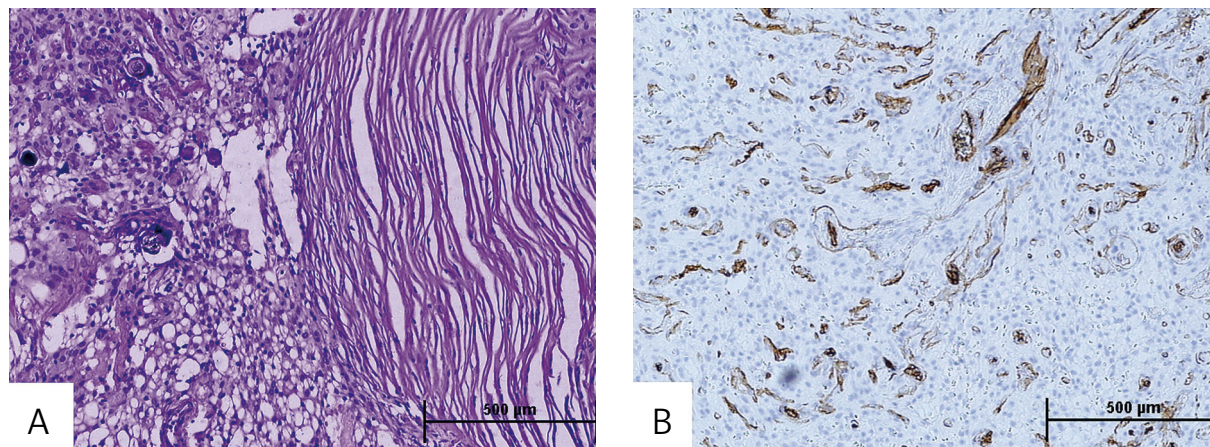


Fig. 3. A) Microscopic images of the tumour, stained with haematoxylin and eosin. Pleomorphic characteristics – psammoma bodies, microcysts and fibrous component are visible. B) Immunohistochemical staining with a slight positive reaction for CD34. No increased neoangiogenesis was noticed. The bar: 500 µm

tor cerebral venous trunks and finally results in chronic subdural haemorrhage. Patients with large tumours occluding the SSS, who did not qualify for or refused surgery, should be carefully monitored because of possibly increased risk for subdural haemorrhage.

Disclosure

Authors report no conflict of interest.

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