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Primary arachnoid cyst – an early postoperative complication after microsurgical resection: a case report and review of literature

Primarna arahnoidna cista – rana postoperativna komplikacija nakon mikrokirurške resekcije: prikaz slučaja i pregled literature

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Summary

Arachnoid cysts (AC) are benign, non-neoplastic fluid-filled malformations of the arachnoid tissue. Approximately 50-65% occur in the middle cranial fossa and predominantly on the left side, followed by retrocerebellar and convexity locations. Tremendous development and usage of cross-sectional imaging modalities suggest a higher prevalence of AC than previously thought. Since large arachnoid cysts express mass effect on surrounding neurovascular structures, a surgical approach is preferred to passive observation. Nevertheless, the symptomatology is frequently subjective and difficult to validate, and the causal link between symptoms and an arachnoid cyst is often dubious. Therefore, the operative indication and the best surgical modality for patients with AC remain controversial. Surgical options include open-craniotomy or endoscopic cyst fenestration, cystoperitoneal, cystosubdural, ventriculoperitoneal shunt insertion, or marsupialization via a craniotomy. The complications of these procedures include subdural hematomas, hygromas, hydrocephalus, cerebral edema, postoperative secondary arachnoid cyst, and, more rarely, remote intraparenchymal or subarachnoidal hemorrhage.

Key words: middle fossa arachnoid cyst, microsurgical resection and fenestration, remote site intraparenchymal and subarachnoidal hemorrhage

Sažetak

Arahnoidalne ciste (AC) su benigne, ne-neoplastične malformacije arahnoidnog tkiva ispunjene tekućinom. Otprilike 50-65% ih se javlja u srednjoj lubanjskoj jami i pretežno na lijevoj strani, a potom retrocerebelarnim i konveksnim lokalizacijama. Ogroman razvoj slikovnih pretraga ukazuje na veću prevalenciju AC nego ranije. Budući da velike AC izražavaju kompresivni efekt na okolne neurovaskularne strukture, kirurški pristup poželjniji je od pasivnog promatranja. Ipak, simptomatologija je često subjektivna i teško ju je utvrditi, a uzročna veza između simptoma i AC često je sumnjiva. Stoga indikacija za operaciju i najbolji kirurški modalitet za bolesnike s AC ostaju kontroverzni. Kirurške opcije uključuju otvorenu kraniotomiju ili endoskopsku fenestraciju ciste, cistoperitonealno, cistosubduralno ili ventrikuloperitonealno plasiranje šanta ili marsupijalizaciju putem kraniotomije. Komplikacije ovih zahvata uključuju subduralne hematome, higrome, hidrocefalus, cerebralni edem, postoperativnu sekundarnu arahnoidnu cistu i rjeđe udaljena intraparenhimalna ili subarahnoidalna krvarenja.

Ključne riječi: arahnoidna cista srednje jame, mikrokirurška resekcija i fenestracija, intraparenhimalno i subarahnoidalno krvarenje na udaljenom mjestu

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Introduction

Arachnoid cysts (AC) are benign congenital malformations first described in 1831. They are formed by the splitting or duplication of the arachnoid membrane allowing a clear fluid, resembling normal cerebrospinal fluid, to fill its space.[1] They can occur also secondary as a complication following trauma, infection, bleeding, or surgical manipulation. In all intracranial space-occupying lesions only 1% fall on them with the prevalence in adults at approximately 1.4% and in children at 2.6%.[2] The clinical presentation of AC is variable from asymptomatic up to nonspecific symptoms such as headache, dizziness, balance impairment, and cognitive or behavioral impairment. The diagnosis is set through cerebrospinal imaging and it reveals a sharply demarcated, non-enhancing, extra-axial cyst, with a density/signal similar to the CSF.[3] Computed tomography (CT) can be helpful, but magnetic resonance imaging (MRI) is the gold standard as a definitive diagnostic tool for the evaluation of AC. It shows homogenous T2-weighted signal hyperintensity within the cyst similar to that of CSF which is also confirmed by the fluid attenuated inversion recovery sequences (FLAIR). Galassi proposed a classification scheme for AC based on their communication with the adjacent cisterns on CT scan into three basic types: type I (small, spindle-shaped; limited to the anterior portion of the middle cranial fossa (MCF); free communication of subarachnoid space); type II (superior extent along Sylvian fissure; displacement of the temporal lobe; slow communication with subarachnoid space); type III (large, fills the whole MCF; displacement of temporal, frontal and parietal lobes, little communication with subarachnoid space (Figure 1).[4]

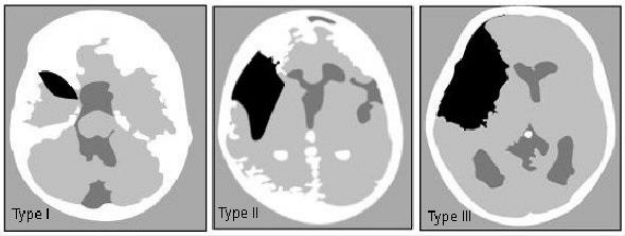

Figure 1 CT classification of Sylvian fissure arachnoid cysts[5]
Slika 1. CT klasifikacija arahnoidnih cisti Silvijeve fisure[5]

Controversy still exists about the best treatment modality for intracranial AC. There are 3 major surgical methods: craniotomy (opening or micro resection) with marsupialization into the subarachnoid spaces, basal cisterns or ventricles; neuro endoscopic fenestration or stereotactic aspiration and shunt surgery (cystoperitoneal, cystosubdural or ventriculoperitoneal).[5] The qualities of each are still a matter of debate. Table 1 shows data from the largest surgical series published in the past 10 years. The complications of these procedures include subdural hematomas, hygromas, hydrocephalus, cerebral edema, postoperative secondary AC**,** and more rarely intraparenchymal or subarachnoidal hemorrhage.[6]

We report large, unilateral MCF AC (Gallasi score III) linked with mild undefined symptoms and treated surgically via micro resection and fenestration of cyst walls followed by remote intraparenchymal and subarachnoid hemorrhage as an initial early postoperative complication.

Table 1 Largest surgical series in the past ten years with results of retrospective studies
Tablica 1. Najveće kirurške serije u posljednjih deset godina s rezultatima retrospektivnih studija

Largest surgical series *Najveće kirurške serije*	No pts *Broj bol*	OSG	EG	SG	OSG +SG
Different Surgical Options for the Management of Intracranial Arachnoid Cysts: retrospective study (2011-2019) *Različite kirurške opcije za liječenje intrakranijalnih arahnoidnih cista: retrospektivna studija (2011.-2019.)* Kayhan, Sait, and Adem Doğan (2022)	44	34		2	8
Results of surgical treatment in patients with intracranial arachnoidal cysts: retrospective study (2015-2019), single center experience *Rezultati kirurškog liječenja bolesnika s intrakranijalnim arahnoidnim cistama: retrospektivna studija (2015.-2019.), iskustvo jednog centra* Masoudi, Mohammadsadegh, Omid Yousefi, and Pouria Azami (2021)	29	26	2	1	
Experience with Management of Intracranial Arachnoid Cysts: retrospective observational study (2004-2020)	56	10	35	11	

Largest surgical series *Najveće kirurške serije*	No pts *Broj bol*	OSG	EG	SG	OSG +SG
Iskustvo s liječenjem intrakranijalnih arahnoidnih cista: retrospektivna opservacijska studija (2004.-2020.) Deopujari, Chandrashekhar E., et al. (2021)					
Surgical management of brain arachnoid cysts: retrospective study (2014-2019) *Kirurško liječenje arahnoidnih cista mozga: retrospektivna studija (2014.-2019.)* Aljubour, Raed M., et al. (2022)	65	18	23	24	
Comparison of Surgical Techniques for Intracranial Arachnoid Cysts: A Volumetric Analysis (2010-2020) *Usporedba kirurških tehnika za intrakranijalne arahnoidne ciste: volumetrijska analiza (2010.-2020.)* Kirmizigoz, Sahin, et al. (2023)	66	32	17	11	6

No pts- number of patients / *broj bolesnika*, OSG- Open surgical group /*otvorena kirurška grupa*, EG- Endoscopic group/ *endoskopska grupa*, SG- Shunt group / *šant grupa*, OSG+ SG- Open surgical group+ surgical group / *otvorena kirurška grupa + kirurška grupa*

Case report

A 44-year-old female Caucasian presented with a 2-year history of blunt, intermittent headache accompanied by anxiety and impaired verbalization. Several times examined and treated by a family practitioner and neuropsychiatrist with partial alleviation of problems. Since ailments did not pass, she was referred to a neurosurgeon. On admission, she was Glasgow Coma Score 15 with no signs of focal neurological deficit. Laboratory tests had referent values. MRI revealed right Gallasi III stage frontotemporal AC (Figure 2).

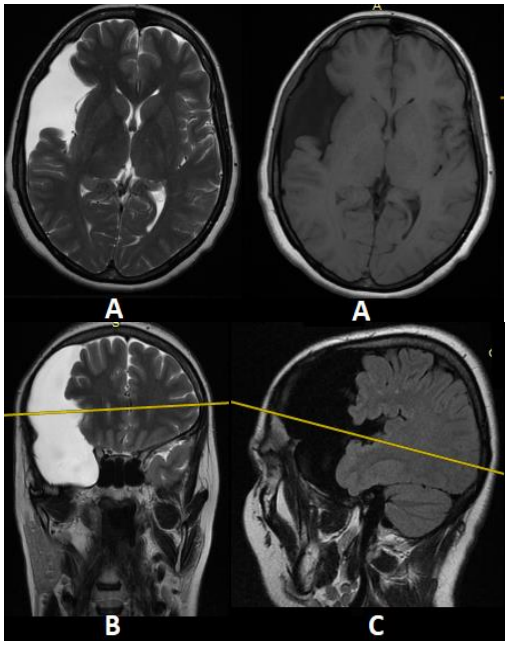

Figure 2 Axial T2, T1(A), coronal T2 (B), and sagittal T1 (C) non-contrast-enhanced magnetic resonance imaging (MRI) indicates a large right frontotemporal arachnoid cyst (Department of Radiology, Zenica Cantonal Hospital, 2020)

Slika 2. Aksijalna T2, T1(A), koronalna T2 (B) i sagitalna T1 (C) sekvenca magnetne rezonance (MRI) bez kontrasta ukazuje na veliku desnu frontotemporalnu arahnoidnu cistu (Odjel za radiologiju, Kantonalna bolnica Zenica, 2020.)

Surgical treatment was considered as one of the modalities of treatment. After a large right-sided open craniotomy and durotomy, cystic wall with a tiny fibrous wall was observed. Initial fenestration of the cyst showed the bright fluid under high pressure. Sylvian and prepontine cisterns were fenestrated after micro resection of the cystic wall in order to establish communication with the subarachnoid spaces in several places. A histopathological specimen showed classical features of delicate cystic structure (Figure 3).

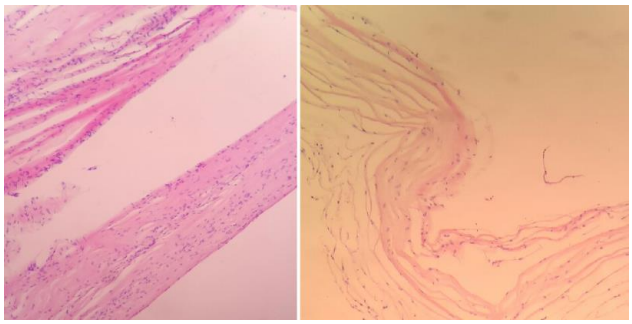

Figure 3 Microphotography of a pathohistological section showing histological features of a delicate cystic structure lined by a flattened or cuboidal epithelium. Cystic wall stained with hematoxylin and eosin (HE), original magnification of × 10 (Department of Pathology, Zenica Cantonal Hospital, 2020)

Slika 3. Mikrofotografija patohistološkog presjeka koji pokazuje histološke karakteristike delikatne cistične strukture obložene spljoštenim ili kockastim epitelom. Cistični zid obojen hematoksilinom i eozinom (HE), originalno uvećanje × 10 (Odjel za patologiju, Kantonalna bolnica Zenica, 2020.)

Due to aggravated awakening during the early postoperative period, urgent head CT imaging was performed, which revealed signs of ex vacuo supra et infratentorial subarachnoid hemorrhage linked with intraparenchymal hemorrhage and 8mm midline shift (Figure 4) after which the patient was retained in the intensive care unit (ICU).

For the next three days, the patient was cardiorespiratory stable under neuro intensivist monitoring. On the 5th postoperative day, the patient gradually woke up, sluggish, with no signs of lateralization, satisfactory respiratory and pupil status and aggravated verbalization. Since the clinical-neurological status was gradually improving and control head CTs were satisfactory, after a few days, the patient was transferred to the parent department and after that, to the regional rehabilitation center for further treatment in a solid physical condition without clear signs of neurological deficit. On a regular control, one year after discharge, the patient's general condition was better, a neurological condition satisfying and radiologically with signs of refilling at the site of the prior cystic cavity (Figure 5).

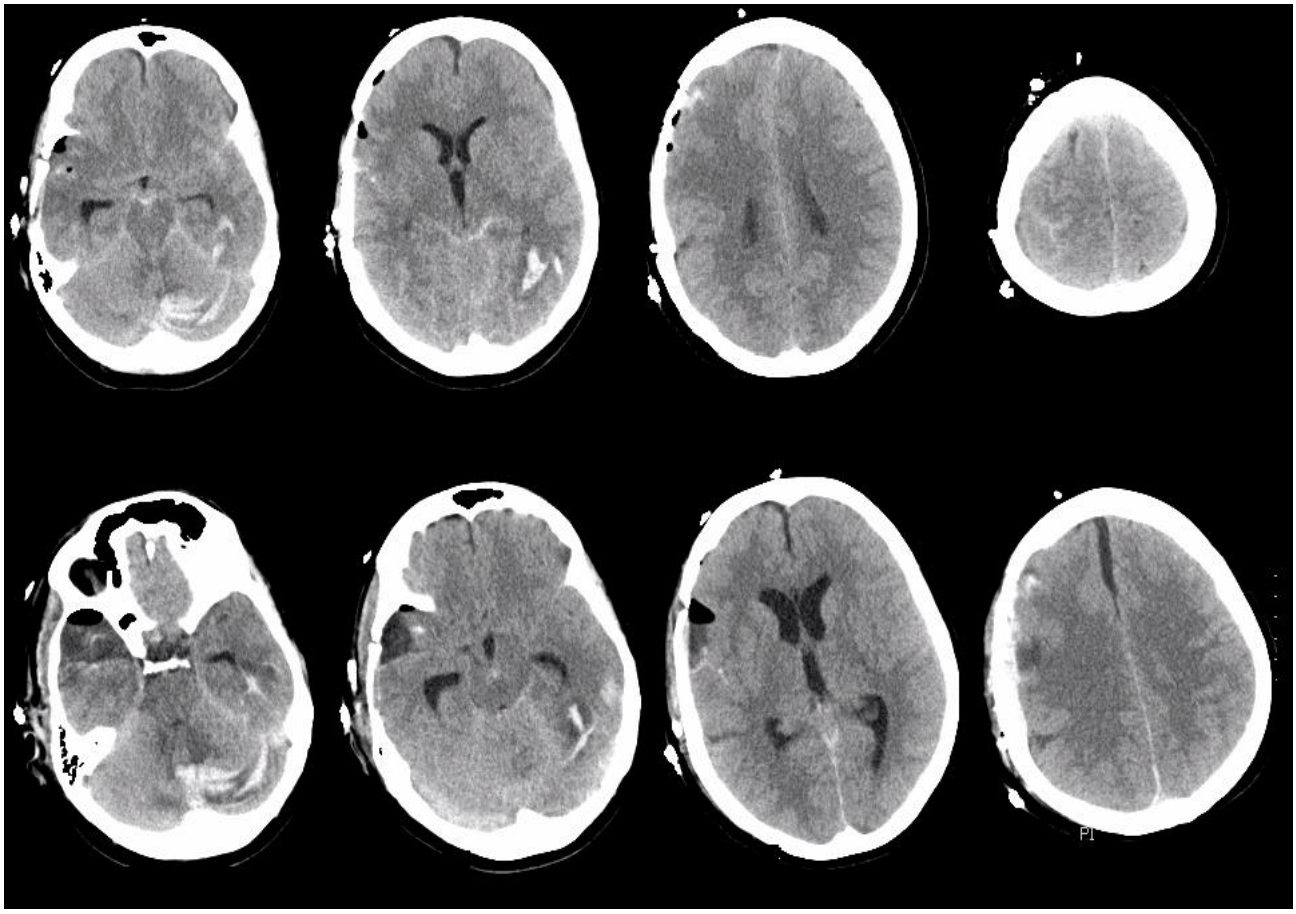

Figure 4 Postoperative axial non-contrast-enhanced computed tomography (CT) shows signs of supra and infratentorial subarachnoidal hemorrhages linked with left temporooccipital intraparenchymal hematoma (Department of Radiology, Clinical Center University of Sarajevo 2020.)

Slika 4. Postoperativna aksijalna kompjuterizovana tomografija (CT) bez kontrasta pokazuje znakove supra i infratentorijalnog subarahnoidalnog krvarenja povezanog sa lijevim temporookcipitalnim intraparenhimalnim hematomom (Odjel za radiologiju, Kantonalna bolnica Zenica, 2020.)

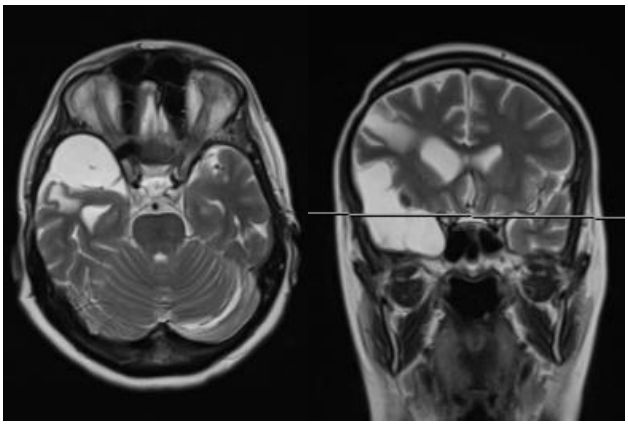

Figure 5 Axial (T2) and coronal (T2) non-contrast-enhanced magnetic resonance imaging (MRI) indicates refilling at the site of the prior cystic cavity one year later (Department of Radiology, Zenica Cantonal Hospital, 2020).
Slika 5. Aksijalna (T2) i koronalna (T2) magnetna rezonanca (MRI) bez kontrasta ukazuje na ponovno punjenje na mjestu prethodne cistične lezije godinu dana kasnije (Odjel za radiologiju, Kantonalna bolnica Zenica, 2020.)

Discussion

Arachnoid cysts are benign, extra parenchymal, intra arachnoidal collections of fluid developed by splitting or duplication of the arachnoidal membrane. They exhibit a male predominance with a 3:1 ratio within the left cerebral hemisphere which did not confirm our case.[7] It can be located anywhere with preponderance in the MCF (50%), usually unilateral and single.[8]

Our case study presents a middle-age female Caucasian with large right-sided MCF AC whose symptoms are thought to be the result of long-term pressure on surrounding brain tissue, changes in CSF dynamics, or dysgenesis of the brain. Two theories describe the pathogenesis of AC. Robinson's theory proposes primary temporal lobe agenesis, while Starkman proposes the AC as the primary abnormality leading to eventual temporal lobe hypoplasia secondary to cyst expansion.[9]

Due to the more widespread use of cross-sectional imaging modalities, the number of newly discovered patients with AC has increased significantly. Morris et al., through a meta–analysis of 16 studies of incidental brain MRI, concluded that AC are the single most prevalent incidental finding, which did not confirm our case.[10]

The lack of evidence-based data has led to considerable controversy regarding the appropriate treatment protocol for AC which mostly remain asymptomatic over time. After all, it is not questionable what to do with small asymptomatic or larger symptomatic arachnoid cysts. The most challenging treatment decision is to treat patients with a medium-sized arachnoid cyst and mild, undefined symptoms when it is unclear whether the symptoms are causally related to the presence of the cyst.

Since our patient had mild, undefined symptoms, we believed that the symptoms were the result of cyst enlargement and consecutive pressure to the brain parenchyma, leading to a combination of symptoms. Controversy still exists about the best treatment modality for intracranial AC mentioned above in the introduction. Open surgery with complete excision of the cyst membrane seems to be the logical treatment modality by most neurosurgeons, but it is unfortunately rarely performed because of the close anatomical relationship between the membrane and the underlying neural tissue.

A prospective study from Rabiei Katrin et al. concluded that fenestration/resection surgery is controversial in the absence of cerebrospinal fluid-pathway blockage and objective signs of clinical improvement.[11] Another prospective, population-based study from Rabiei Katrin et al. speaks against objectively verifiable improvement following surgical treatment in adults with intracranial AC and that it does not justify surgical treatment.[12]

Despite the results of these studies and as surgical treatment is partly a subjective decision, we decided to perform cyst resection with multiple fenestrations toward cisternal spaces and as a result of the sudden decompression we had signs of ex vacuo remote subarachnoid and intraparenchymal hemorrhage.

The rapid decompression with the craniotomy produces a rapid rise in cerebral perfusion, and changes in the intracranial dynamics with venous hyperemia resulting in consecutive parenchymal hemorrhage distant from the site of operation due to "shift of the brain". This is the explanation for intraparenchymal hemorrhage in the report of Živković N. et al.[13] Bahl Anuj et al. have shown that intraparenchymal hemorrhage in remote areas following the evacuation of a large AC indicates that chronic high cerebral perfusion pressure exists and there is a poor local auto-regulatory response surrounding an AC. It has been postulated that this complication probably represents a reperfusion injury similar to the normal perfusion pressure breakthrough syndrome following surgery for arterio-venous malformations.[14] Published data from the last decade, 2013-2022, revealed only one case of remote intraparenchymal hemorrhage after fenestration of an intracranial arachnoid cyst.[15]

Peter L. et al. confirmed the thesis that if the clinical-neurological status is not convincing, the

resection of the giant AC can lead to a vicious circle without a final solution. They described the enlargement of the ventricles and CT signal changes consistent with periventricular edema.[16]

Conclusion

Our report confirms that, given the potential of serious postoperative complications, surgical treatment should be indicated in a very narrow range of clinically symptomatic patients taking into account the previous medical history, localization, and cyst behavior and the surgeon's familiarity with all possible surgical procedures and potential complications. This statement is particularly convincing if you consider that the cystic lesion returned one year later, which ultimately leaves doubt on the correctness of the assessment and justification of the mentioned treatment.

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