

Infrequent case of cavum septi pellucidi empyema and principles of neurosurgical management: case report and literature review

Rijetki slučaj empijema cavum septi pellucidi i principi neurokirurškog zbrinjavanja: prikaz slučaja s pregledom literature

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Summary

Aim: To present a very rare case of empyema cavuma septi pellucidi.

Case report: A 5-year-old male child was admitted to the Department of Infectious Diseases Cantonal Hospital Zenica because of fever (38.30°C), headache and vomiting. The patient developed intracranial hypertension as a result of a compressive purulent collection formed due to meningitis between the laminae of the septum pellucidum with consequent intracranial hypertension.

Conclusion: The decision regarding the modality of treatment was not easy. We considered that empyema evacuation using the transcalsalinterhemispheric approach allows the complete removal of purulent collection and the placement of drainage, which allows additional emptying of the empyema cavity and prevents empyema recurrence. Empyema evacuation with drainage and antibiotic therapy have shown beneficial results.

Key words: purulent content, septum pellucidum, neurosurgery

Sažetak

Septum pelucidum je tanka, dvoslojna pregrada situirana između korpuska lozuma i forniksa. Najčešća anatomska varijacija je perzistentan kavum septi pelucidi. Nakupljanje gnojnog sadržaja između lamina septum peluciduma je vrlo rijetko. Opisano je samo nekoliko slučajeva u literaturi. Prezentirali smo zanimljiv slučaj djeteta koje je razvilo intrakranijalnu hipertenziju kao rezultat kompresije gnojne kolekcije između lamina septum peluciduma, a koja je nastala kao komplikacija meningitisa. Evakuacija empijema i antibiotska terapija dali su dobre rezultate.

Ključne riječi: gnojni sadržaj, septum pelucidum, operacija

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Introduction

The septum pellucidum is a thin two-layered septum situated between the corpus callosum and the

fornix. It consists of glial cells, scattered neurons and fibers.¹ The ventricular surface of the septum is covered with ependyma. The septum pellucidum is connected with the hippocampus and hypothalamus,

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but its function is not completely clear. It is subject to anatomical variations.^{1,2} According to various studies, the incidence of cavum septipellucidi ranges from 0.7% to 37%.³ Inflammatory process and accumulation of purulent content between the lamina septum pellucidum is very rare. Only a few cases have been described in the literature.⁴

The following is an interesting case of a child who developed intracranial hypertension as a result of a compressive purulent collection between the laminae of the septum pellucidum formed due to meningitis.

Case report

A 5-year-old male child was admitted to the Department of Infectious Diseases Cantonal Hospital Zenica because of fever (38.3°C), headache and vomiting. The symptoms had started three days prior to admission. His neck was stiff, Kernig's sign was positive, but there was no neurological deficit. Laboratory tests showed inflammation: WBC $17.35 \times 10^9/L$; RBC $3.70 \times 10^{12}/L$; Hgb 93.6 g/dL; Hct 0.30 L/L; MCV 80.3 fL; MCH 25.3 pg; MCHC 31.5 g/dL; RDW 11.2%CV; PLT $320 \times 10^9/L$; MPV 5.9 fL; Glucose 4.7 mmol/L; Urea 2.8 mmol/L; Creatinine 22 $\mu\text{mol}/L$; AST 31 U/L; ALT 18 U/L; fibrinogen 6.79 g/L; CRP 0.090 g/L. The analysis of the sampled cloudy cerebrospinal fluid (CSF) showed 850 white cells (63% neutrophils), protein concentration 0.5 g/L, and the level of glucose 3.1 mmol/L. The bacteriological analyses of CSF, haemoculture and urinculture were negative. Other diagnostic tests were normal. Due to clinical considerations of acute meningitis, the patient was treated with broad spectrum antibiotics and antiedematous therapy. The patient had history of previously diagnosed CSP (Figure 1).

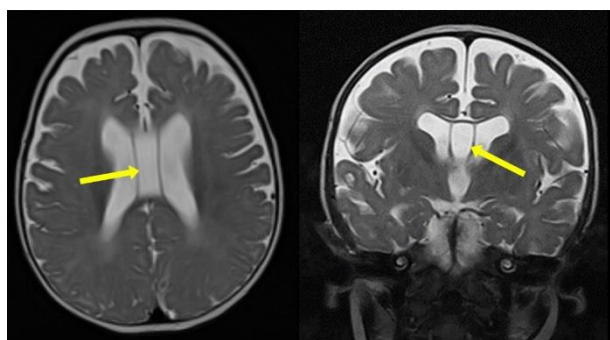


Figure 1 MRI (T2 axial and coronal scans): persistent cavum septipellucidi (yellow arrow); four years before the onset of empyema (Department of Radiology Cantonal Hospital Zenica)

Slika 1. MRI (T2 aksijalni i koronalni presjeci): perzistentni kavu septipellucidi (žuta strelica);

četiri godine prije pojave empijema (Odjel za radiologiju Kantonalne bolnice Zenica)

During hospitalization the patient showed subjective improvement with an occasional increase in body temperature. Control laboratory findings have shown improvement: WBC $12.94 \times 10^9/L$ (Ne 0.59; Ly 0.31); RBC $4.14 \times 10^{12}/L$; Hgb 106.2 g/dL; Hct 0.34 L/L; MCV 81.6 fL; MCH 25.7 pg; MCHC 31.5 g/dL; RDW 12.2%CV; PLT $547 \times 10^9/L$; MPV 4.6 fL; Glucose 6.9 mmol/L; AST 19 U/L; ALT 7 U/L; CRP 54.70 g/L; Na 129 mmol/L, K 5.5 mmol/L, Cl 93 mmol/L. The CSF was clear and contained 41 white cells/ mm^3 with protein concentration 0.8 g/L.

On the twenty-first day of hospitalization, the condition worsened due to the development of severe headache, vomiting and gradual loss of consciousness. An emergency magnetic resonance imaging scan (Siemens Magnetom Avanto 1.5 T, Erlangen, Germany) was performed which showed a massive interhemispheric cystic collection in the area of the septum pellucidum with consequent obstruction of CSF flow (Figure 2).

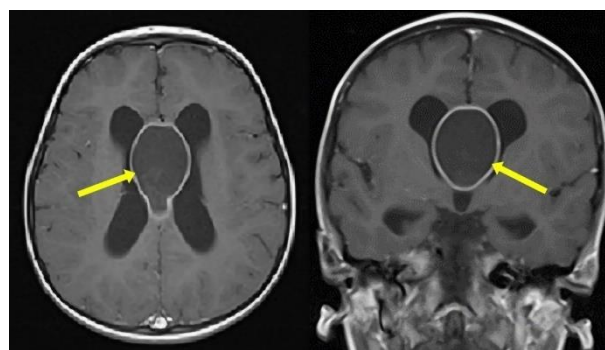


Figure 2 MRI (T2 axial and coronal scans): massive interhemispheric cystic collection (yellow arrow) in the area of the septum pellucidum with consequent obstruction of CSF flow (Department of Radiology Cantonal Hospital Zenica)

Slika 2. MRI (T2 aksijalni i koronalni presjeci): masivna interhemisferična cistična nakupina (žuta strelica) u području septum peluciduma s posljedičnom opstrukcijom cerebrospinalnog protoka (Odjel radiologije Kantonalne bolnice Zenica)

The neurosurgeon indicated emergency microsurgery to remove the pus collection and place drainage. An anterior interhemispheric approach with craniotomy (4×4cm) above the superior sagittal sinus was performed, predominantly right. The dura incision provided an interhemispheric approach, between the falx and the right hemisphere to the corpus callosum. Pericalous arteries were slightly

dissected and a callosotomy was performed to show the empyema capsule. After incision of the capsule and removal of the yellowish - thick collection, the cyst cavity was washed with antibiotic solution, and then a silicone drain was placed (Figure 3).

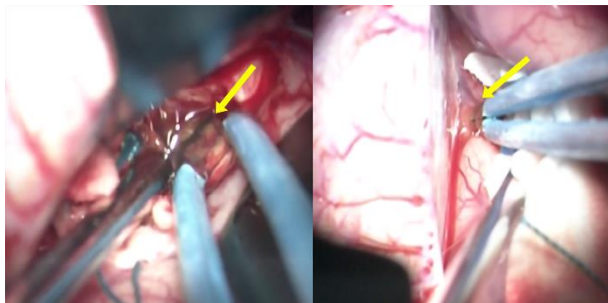


Figure 3 Microsurgically evacuation of CSP empyema using anterior interhemispheric transcalsal approach (yellow arrow) (Department of Neurosurgery Cantonal Hospital Zenica)

Slika 3. Mikrokirurška evakuacija CSP empijema prednjim interhemisferičnim transkaloznim putem žuta (strelica) (Odjel neurokirurgije Kantonalne bolnice Zenica)

Hemostasis is revised, and the dura was sutured directly. The bone flap was fixed with sutures and wound was sutured by layers. The patient was transferred to the Intensive Care Unit (ICU) for postoperative treatment. The patient has shown improvement. The following day, we performed a control Computed Tomography of the brain which showed a correct postoperative finding (Figure 4).



Figure 4 CT (axial non contrast scan) shows a correct postoperative finding with adequate catheter position (yellow arrow). There is an air collection due to the craniotomy on the right frontal area (Department of Radiology Cantonal Hospital Zenica)

Slika 4. CT (aksijalni nekontrastni presjek) pokazuje uredan postoperativni nalaz s adekvatnom pozicijom katetera (žuta strelica). Prisutna je kolekcija zraka zbog kraniotomije u desnom frontalnom području (Odjel za radiologiju Kantonalne bolnice Zenica)

The patient was transferred to the Department of Pediatrics Cantonal Hospital Zenica, and supervised by a neurosurgeon, pediatrician and infectologist. He was treated intravenous with monotherapy of meropenem (3 grams per day) for five weeks. The drain was removed on the 5th day of surgery. The control laboratory findings were correct, and patient was discharged 11 days after surgery. At the control neurosurgical examination the patient had completely normal neurological findings, as well as MRI (Figure 5).

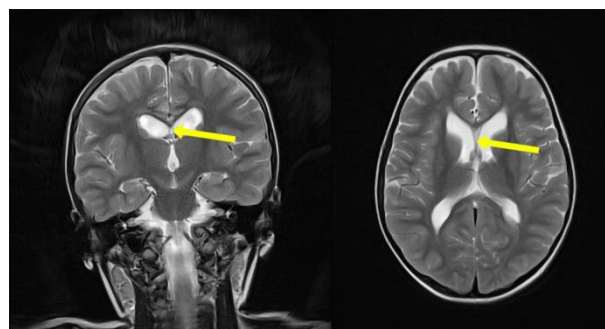


Figure 5 MRI (T2 axial and coronal scans): correct MRI without presence of CSP or empyema (yellow arrow). (Department of Radiology Cantonal Hospital Zenica)

Slika 5. MRI (T2 aksijalni i koronalni presjeci): uredan MRI nalaz bez prisustva CSP i empijema (žuta strelica) (Odjel radiologije Kantonalne bolnice Zenica).

Discussion

The septum pellucidum is a thin two-layered membrane which separates the two lateral ventricles.¹ During the embryonic development three cavities had development between the lateral ventricles. They gradually involuted by the end of the second year of postnatal life.² In some cases cavities remain persistent and cavum septipellucidum (CSP), cavum vergae (CV) and cavum veliinterpositi (CVI) are formed. CSP is part of a normal development. It can be found in all fetuses at 36 weeks gestation and persists in 36% of full-term infants, only 6% of them persist after the six month of life.^{2,3} CSP usually does not cause symptoms and it is an accidental finding. CSP communicates with the lateral and third ventricles. It is filled with CSF.^{1,3}

Empyema of CSP is a very rare clinical condition. Only few cases have been described in the literature.^{4,5} In the described cases, empyema CSP is a complication of meningitis and ventriculitis.⁴⁻⁷

The medication therapy of cerebral empyema includes antibiotic therapy.^{6,7} In cases of intracranial hypertension and worsening of neurological status, surgical evacuation of empyema is indicated.⁵⁻⁷

Looking at the case reports so far (table 1), it can be seen that empyema occurred more often in the male population. The cause, according to López-Pérez et al.,⁸ Mokgokong⁹ and Abe et al.,¹⁰ was head trauma. Li et al.⁶ describes the development of empyema in preterm infants, while Saryyeva et al.¹¹ and Rotman et al.¹² mention previous sinusitis as a possible cause. In this case, empyema of CSP was formed due to meningitis or ventriculitis. The persistent CSP which was diagnosed earlier contributed to the development of empyema. The infection

spread through the CSF to the persistent CSP and led to the formation of a purulent collection. Antibiotic therapy enabled the treatment of meningitis and slowed down the clinical course of the disease causing the manifestation of empyema 21 days after hospitalization.

Previous experience showed the diversity of bacterial agents: *Streptococcus pneumoniae*,^{9,10,12} *Staphylococcus epidermidis*,¹¹ *Acinetobacter baumannii*,⁸ *Proteus mirabilis*⁶ and *Klebsiella pneumoniae*.¹²

Tabele 1 Summary of literature review / *Tablica 1. Sažetak pregleda literature*

References <i>Reference</i>	Patient information <i>Informacije o bolesniku</i>			Cause <i>Uzrok</i>	Bacterial cause <i>Bakterijski uzročnik</i>	Hydrocephalus <i>Hidrocefalus</i>	Treatment <i>Liječenje</i>		Outcome <i>Ishod</i>
	Age <i>Dob</i>	Sex <i>Spol</i>	Anamnesic data <i>Anamnestički podaci</i>				Antibiotics <i>Antibiotici</i>	Surgical <i>Kirurgija</i>	
López-Pérez et al. (8)	37y	M	Head injury with multiple fractures, subarachnoid hemorrhage and pneumocephalus <i>Povreda glave s višestrukim prijelomima, subarahnoidalnim krvarenjem i pneumocefalusom.</i>	Posttraumatic meningitis <i>Posttraumatski meningitis</i>	<i>Acinetobacter baumannii</i>	No <i>Ne</i>	imipenem and colimycin <i>imipenem i kolimicin</i>	Stereotaxic puncture and drainage <i>Stereotaksijska punkcija i drenaža</i>	Lumbar puncture showed normalisation of inflammatory markers, full recovery <i>Lumbalna punkcija pokazala normalizaciju upalnih markera, potpuni oporavak.</i>
Mokgokong S. (9)	20y	M	Head injury (3 years earlier) <i>Povreda glave (3 godine ranije)</i>	Bacterial meningitis <i>Bakterijski meningitis</i>	<i>Streptococcus pneumoniae</i>	Yes <i>Da</i>	penicillin G, chloramphenicol & metronidazole <i>penicillin G, kloramfenikol i metronidazol</i>	Interhemispheric transcallosal approach, aspiration and drainage. <i>Interhemisferični transkalozni pristup, aspiracija i drenaža</i>	CT conformed CSP reduction, without neurological deficiency <i>CT potvrda redukcije CSP, bez neuroloških ispada</i>
Abe et al. (10)	60y	M	Head injury (4 days earlier) <i>Povreda glave (4 dana ranije)</i>	Posttraumatic meningitis <i>Posttraumatski meningitis</i>	<i>Streptococcus pneumoniae</i>	Yes <i>Da</i>	unknown <i>nepoznato</i>	Stereotactic puncture and aspiration of pus. <i>Stereotaksijska punkcija aspiracija gnoja</i>	CT conformed CSP reduction <i>CT potvrda redukcije CSP</i>

References <i>Reference</i>	Patient information <i>Informacije o bolesniku</i>			Cause <i>Uzrok</i>	Bacterial cause <i>Bakterijski uzročnik</i>	Hydrocephalus <i>Hidrocefalus</i>	Treatment <i>Liječenje</i>		Outcome <i>Ishod</i>
	Age <i>Dob</i>	Sex <i>Spol</i>	Anamnestic data <i>Anamnestički podaci</i>				Antibiotics <i>Antibiotici</i>	Surgical <i>Kirurgija</i>	
Li et al. (6)	1m	M	Premature birth and respiratory distress <i>Prijevremeni porod i respiratorni distres</i>	Bacterial meningitis <i>Bakterijski meningitis</i>	<i>Proteus mirabilis</i>	Yes <i>Da</i>	ampicillin and cefotaxime <i>ampicilin i cefotaksim</i>	No <i>Ne</i>	Ultrasonography showed CSP reduction <i>Ultrazvuk pokazao CSP redukciju</i>
Saryyeva et al. (11)	38y	F	Sinusitis (sphenoid sinus) <i>Sinusitis (sfenoidalni sinus)</i>	Suspected meningitis <i>Sumnjana meningitis</i>	<i>Staphylococcus epidermidis & Streptococcus pneumoniae</i>	Yes <i>Da</i>	lindamycin and ceftriaxone <i>lindamicin i ceftriakson</i>	Stereotaxic puncture and drainage. <i>Stereotaksijska punkcija i drenaža</i>	3 days after puncture MRI showed resolution of empyema <i>Tri dana nakon punkcije MRI pokazao rezoluciju empijema</i>
Rotman et al. (12)	51y	M	Chronic sinusitis and sinus surgical procedure <i>Kronični sinusitis i kirurška procedura sinusa</i>	Pansinusitis, otomastoiditis and bacterial meningitis <i>Pansinusitis, otomastoiditis i bakterijski meningitis</i>	<i>Klebsiella pneumoniae</i>	No <i>Ne</i>	ceftriaxone and ciprofloxacinotic drops <i>ceftriakson i ciprofloksacinske kapi za uši</i>	No <i>Ne</i>	Near complete resolution of empyema (MRI). <i>Gotovo kompletna rezolucija empijema (MRI).</i>
Efendić et al. (This study) <i>Ova studija</i>	5y	M	Bacterial meningitis <i>Bakterijski meningitis</i>	Bacterial meningitis <i>Bakterijski meningitis</i>	None isolated <i>Nije izoliran</i>	No <i>Ne</i>	meropenem <i>meropenem</i>	Interhemispheric transcallosal approach, aspiration and drainage. <i>Interhemisferični transkalozni pristup, aspiracija i drenaža.</i>	Control MRI showed resolution of empyema, without neurological deficiency <i>Kontrolni MRI prikazao rezoluciju empijema, bez neuroloških ispada</i>

Legend: y – years; d – days; m - months

Legenda: y – godine; d – dani; m – mjeseci

There is a noticeable difference in the approach to antibiotic therapy. The method of administration in all cases was intravenous, and López-Pérez et al.⁸ opted for imipenem and colimycin. Mokgokong⁹ mentioned penicillin G, chloramphenicol & metronidazole as antibiotic therapy. Li et al.⁶ state that antibiotic therapy included ampicillin and cefotaxime, while Saryyeva et al.¹¹ prefer lindamycin and ceftriaxone. Ceftriaxone in combination with ciprofloxacin ear drops, due to accompanying otitis, was the therapy of choice in Rotman et al.¹² In our case, the therapy of choice was meropenem with intravenous administration for five weeks, which in earlier studies proved to be a good form of monotherapy management of brain abscess.^{13, 14}

Increase of intracranial pressure value and deterioration of the neurological status indicate surgical intervention. Surgical treatment of empyema, along with antibiotic therapy, is the treatment of choice, and it has been proven to lead to a rapid improvement in the patient's neurological status.¹¹ There are different modalities of surgical treatment, as can be seen in table 1. Today, stereotaxic puncture and drainage of CSP empyema is a common treatment modality due to less possibility of developing complications. Mokgokong⁹ chose the interhemispheric transcallosal approach, aspiration and drainage, which was the choice in our case report. Although there are risks of complications such as subarachnoid bleeding of pericallosal blood vessels, and the possible secondary development of ventriculitis due to the spread of pus, this surgical approach gives excellent results. The reason for this is better visualization of the cavity and detailed aspiration of the contents. Hydrocephalus as a possible complication appeared in four case reports.⁹⁻¹¹ In our case, hydrocephalus did not develop.

Empyema evacuation using the transcallosal interhemispheric approach allows the complete removal of purulent collection and the placement of drainage, which allows additional emptying of the empyema cavity and prevents empyema recurrence. Antibiotic therapy involves intravenous administration for four to six weeks¹² with inflammatory parameter monitoring (Leukocytes, CRP, PCT, IL-6).

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