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CASE REPORT

Septic Shock After Surgery in Arnold-Chiari Malformation Type I: A Case Report

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Background: Arnold-Chiari Malformation Type I (ACM-I) is a congenital disorder that can lead to severe neurological symptoms. While decompression surgery is the standard treatment, postoperative complications such as cerebrospinal fluid (CSF) leakage and infections can result in critical outcomes. Here, we report a case of septic shock following decompression surgery in a patient with ACM-I, emphasizing the challenges in postoperative critical care management.

Case Presentation: A 45-year-old woman with rheumatoid arthritis and progressive neurological symptoms underwent decompression surgery for ACM-I. On postoperative day five, CSF leakage was noted at the surgical site, accompanied by fever and leukocytosis. Despite broad-spectrum antibiotics, the patient developed septic shock, requiring mechanical ventilation and vasopressor support. CSF cultures revealed *Acinetobacter baumannii* infection, necessitating surgical debridement and intrathecal colistin. Despite aggressive management, the patient succumbed to septic shock.

Conclusion: This case highlights the critical importance of early detection and aggressive management of postoperative infections in neurosurgical patients. The occurrence of CSF leakage and subsequent septic shock underscores the need for meticulous postoperative monitoring to prevent fatal complications. Timely intervention, including early microbiological assessment and individualized antibiotic therapy, is essential for improving outcomes in high-risk patients.

Keywords: Arnold-Chiari Malformation Type I, septic shock, postoperative complications, rheumatoid arthritis, case report

Background

Arnold-Chiari Malformation Type I (ACM-I) is a congenital neurological disorder characterized by the downward displacement of cerebellar tonsils through the foramen magnum, leading to cerebrospinal fluid (CSF) flow obstruction and subsequent neurological deficits.[1] While decompression surgery is the standard treatment for symptomatic ACM-I, it carries significant postoperative risks, particularly CSF leakage and infectious complications, which can result in life-threatening conditions such as septic shock.[2]

One of the critical concerns in postoperative care for ACM-I patients is early recognition and management of complications, particularly in individuals with underlying conditions that may impair immune response.[3] Patients with rheumatoid arthritis (RA), such as the case presented here, are inherently at a higher risk for infections due to chronic inflammation and immunosuppressive therapies (eg, corticosteroids and methotrexate).[4] These factors can complicate postoperative healing and increase susceptibility to severe infections, making meticulous monitoring in the intensive care unit (ICU) essential.

This case report discusses the rapid progression from CSF leakage to septic shock in an ACM-I patient, emphasizing the importance of timely microbiological assessment, aggressive infection control, and individualized antibiotic therapy in critical care settings. By examining the challenges encountered in managing this patient, we aim to provide insights into improving postoperative surveillance and early intervention strategies in neurosurgical patients at high risk of sepsis.

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Case Presentation

The patient is a 45-year-old married woman with a known history of rheumatoid arthritis for about ten years, which was being managed with medication. She reported no other medical conditions in her medical history. In terms of medication, she had been taking NSAIDs such as ibuprofen and naproxen to manage her pain. At times, under her physician's guidance, she also took methylprednisolone and methotrexate periodically to control her condition. Recently, she presented to medical facilities with complaints of neck pain radiating to her arms, along with numbness and weakness in the upper limbs. These symptoms had started about a year prior and had intensified in recent months, not responding to prior treatments. During physical examinations, the patient showed muscle weakness in the upper limbs but had no other symptoms, such as gait disturbance, imbalance, headache, or dizziness. In neurological examinations, muscle strength was recorded as 3–4 out of 5 in the right upper limb, 2–3 out of 5 in the left upper limb, and 5 out of 5 in both lower limbs. Initial head and neck X-Ray was obtained (Figure 1).

A brain MRI of the patient (shown in Figure 2) was used for further evaluation and differential diagnosis. Based on the image, the extent of tonsillar herniation is estimated to be approximately 8 to 10 millimeters below the foramen magnum, which is consistent with the diagnostic criteria for Chiari Malformation Type I (defined as herniation greater than 5 millimeters).

Based on imaging results, a diagnosis of ACM-I was made. Following this diagnosis, the patient underwent posterior fossa decompression surgery, which included decompressive craniectomy, resection of the foramen magnum, C1

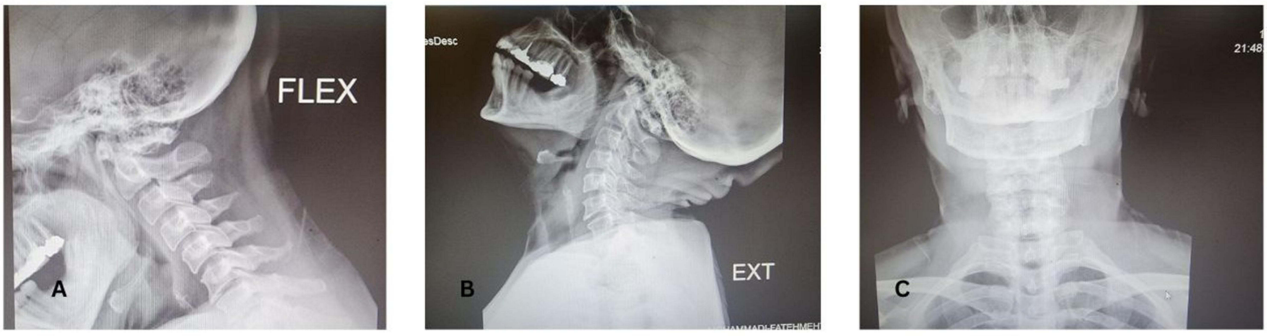

Figure 1 Radiograph of the cervical spine (**A**) Flexion lateral view and (**B**) Extension lateral view (**C**) anterior-posterior view. The cervical vertebrae are normal and free of fractures and dislocations.

Figure 2 MRI of the brain without contrast of Arnold-Chiari malformation I. The sagittal view demonstrates the herniated cerebellar tonsils descending below the level of the foramen magnum.

laminectomy, V-shaped durotomy, coagulation of the tonsils, and duraplasty. Part of the timeline of events changes is provided in Table 1.

After surgery, she was transferred to the intensive care unit for continued treatment. In addition to postoperative supportive care, she was started on antibiotics: ceftazidime 2 grams every 8 hours and vancomycin 1 gram every 12 hours. According to the neurosurgery team's recommendations, the patient's head was maintained in an elevated position during the ICU stay to facilitate venous drainage and minimize the risk of cerebrospinal fluid (CSF) leakage. In addition, lactulose syrup was prescribed as a stool softener to prevent constipation and reduce the likelihood of excessive straining during defecation, thereby minimizing sudden increases in intra-abdominal and intracranial pressures.

On the sixth day of hospitalization, CSF leakage was observed at the surgical site, and the patient developed a fever. Despite receiving broad-spectrum antibiotic treatment, her condition worsened by the sixth day, with a significant increase in white blood cell count (WBC: 15,800). At this point, ceftazidime was discontinued, and meropenem 1 gram every 8 hours was started to control the infection. However, despite these adjustments, her WBC count reached 38,600 by the eleventh day. Part of the timeline of laboratory changes is provided in Table 2.

Table 1 Timeline of Events and Daily Vital Signs of the Patient During the Hospitalization

Day	Clinical Event	Laboratory Findings	Management	Vital Signs
Day 1	Admission due to worsening neurological symptoms	Normal CBC, CRP 4.5 mg/L	MRI confirmed ACM-I; decision for surgery	BT (°C): **36.8** BP (mmHg): **110/70** HR (bpm): **84** RR (/min): **18** SpO_2 (%): **98**
Day 2	Posterior fossa decompression surgery	-	ICU admission, prophylactic antibiotics (ceftazidime + vancomycin)	BT (°C): **36.9** BP (mmHg): **108/68** HR (bpm): **86** RR (/min): **18** SpO_2 (%): **98**
Day 6	CSF leakage detected at surgical site, fever (38.9°C), WBC (15,800)	CSF WBC 2500, Protein 77 mg/dL	Antibiotic escalation to meropenem	BT (°C): **38.9** BP (mmHg): **92/58** HR (bpm): **110** RR (/min): **24** SpO_2 (%): **91**
Day 7	Clinical deterioration: hypotension, tachycardia, worsening leukocytosis (WBC 22,500)	CSF culture: Acinetobacter baumannii, CSF WBC 350, CRP 43.2 mg/L	ICU resuscitation, norepinephrine started	BT (°C): **39.1** BP (mmHg): **85/55** HR (bpm): **118** RR (/min): **24** SpO_2 (%): **90**
Day 9	Septic shock, mechanical ventilation required	Blood culture: Negative; Urine culture: Candida albicans	Intrathecal colistin added	BT (°C): **39.6** BP (mmHg): **80/50** HR (bpm): **124** RR (/min): **28** SpO_2 (%): **88**
Day 10–11	Progressive multi-organ dysfunction	Blood culture: Negative, CSF culture: Negative	Caspofungin initiated, but patient deteriorated	BT (°C): **40.0** BP (mmHg): **70/40** HR (bpm): **136** RR (/min): **30** SpO_2 (%): **86**

Abbreviations: CBC, Complete Blood Count; CSF, Cerebrospinal Fluid; CRP, C-reactive protein; WBC, White Blood Count; MRI, Magnetic Resonance Imaging; BT, Body Temperature; BP, Blood Pressure; HR, Heart Rate; RR, Respiratory Rate.

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Table 2 A Timeline of Some of the Patient's Laboratory Changes

Hospitalization Days	CBC Test			CSF Analysis Test					Cultures Test				CRP
	WBC	Segment	Lymphocyte	WBC	RBC	Glucose	Protein	LDH CSF	B/C	U/C	Catheter	CSF	4.5
Day 1	8.7	64.5%	25%	–	–	–	–	–	–	–	–	–	–
Day 2	14	80%	11.5%	–	–	–	–	–	–	–	–	–	–
Day 3	16.1	89%	6.6%	–	–	–	–	–	–	–	–	–	–
Day 4	10.7	80%	11.2%	–	–	–	–	–	–	–	–	–	–
Day 5	11.3	84.7%	7.9%	–	–	–	–	–	–	–	–	–	–
Day 6	15.8	87%	4.6%	2500	750	77	4	8	–	–	–	–	–
Day 7	22.5	87%	3%	350	500	60	82	10	–	–	–	Acinetobacter baumannii	293
Day 8	19.6	92%	2.4%	7600	200	3	266	12	–	–	–	–	–
Day 9	17.9	96%	2.5%	–	–	–	–	–	No Growth	Candida albicans	–	–	–
Day 10	14.6	90%	4.7%	0	750	86	14	206	No Growth	–	–	–	–
Day 11	38.6	93.6%	3.2%	–	–	–	–	–	No Growth	–	No Growth	No Growth	–

Abbreviations: CBC, Complete Blood Count; CSF, Cerebrospinal Fluid; CRP, C-reactive protein; WBC, White Blood Count; RBC, Red Blood Cell; LDH, Lactate Dehydrogenase; B/C, Blood Culture Test; U/C, Urine Culture.

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After the laboratory changes, the patient's hemodynamic status became unstable, leading to the need for intubation to provide cardiopulmonary support. On the ninth day, the patient was taken back to the operating room for extensive debridement and washing of the surgical site. Additionally, CSF culture results indicated infection with Acinetobacter baumannii, which is sensitive to colistin. Based on the CSF culture report, infectious disease, and clinical pharmacology consultations were carried out. Following these consultations, the patient's previous antibiotics were discontinued, and broad-spectrum antibiotics were started, including intravenous and intrathecal colistin and intravenous ampicillin-sulbactam 3 grams every 6 hours. Due to the positive Candida culture from the urinary catheter, the patient's Foley catheter was replaced. On day 7 of hospitalization, the patient developed septic shock with persistent hypotension (BP 85/55 mmHg, HR 118 bpm, SpO_2 90%). Norepinephrine infusion was started at 0.1 μg/kg/min and was gradually titrated to 0.5 μg/kg/min by day 9 in an effort to maintain MAP ≥65 mmHg. However, the patient's condition remained unstable despite maximal vasopressor support. Additionally, the antifungal drug caspofungin 75 mg was administered as a bolus and gradually increased to 50 mg daily. Despite maximal supportive care in the intensive care unit, including broad-spectrum antibiotics and high-dose norepinephrine infusion, the patient's condition deteriorated and she ultimately died on hospital day 11 due to refractory septic shock.

Discussion and Conclusion

This case underscores the importance of proper and timely management of postoperative complications in patients with ACM-I. Although posterior fossa decompression surgery is considered the standard treatment for these patients and can lead to symptom improvement, complications such as CSF leakage and severe infections can result in adverse outcomes. In this case, despite aggressive medical and supportive management in the intensive care unit, the patient's condition rapidly deteriorated, ultimately leading to death from refractory septic shock.

A review of the medical literature reveals that ACM-I, particularly after posterior fossa decompression surgery, can be associated with postoperative complications such as meningitis, abscess, and septic shock. For instance, the results of our study are in significant agreement with previous research findings. A survey by Passias et al[5] showed that patients with ACM-I who underwent surgery faced substantial risks, including postoperative complications such as neurological issues, anemia, and respiratory distress. This study also indicated that patients who underwent spinal fusion, particularly those with fusion levels of four or more, had a significantly higher incidence of complications. These findings align with the results of our study, as our patient also experienced severe complications after surgery. This study emphasizes the importance of postoperative care and careful monitoring of patient's conditions.

Additionally, the results of other studies have shown that comorbidities such as cardiovascular, pulmonary, renal diseases, and malignancies can increase the risk of postoperative complications in these patients.[5–7] In our study, underlying rheumatoid arthritis and other potential predisposing factors may have contributed to the increased surgical risk. Therefore, it is recommended that future studies examine the impact of underlying conditions on surgical outcomes. This highlights the importance of comprehensive patient evaluation and management of these conditions before surgery, which can help reduce postoperative complications.

Meticulous attention to the quality of dural closure is a critical step in preventing postoperative cerebrospinal fluid (CSF) leakage and its potentially severe complications, such as meningitis and septic shock. To minimize the risk of postoperative CSF leakage following posterior fossa decompression, several surgical and postoperative strategies have been recommended in the literature. These include performing an intraoperative Valsalva maneuver to identify occult dural tears, avoiding full-thickness dural coagulation to preserve dural integrity, meticulous multilayer dural closure with watertight suturing techniques, and the use of graft augmentation when necessary.[2] Postoperatively, maintaining appropriate head elevation, minimizing excessive straining or coughing, and close monitoring for early signs of leakage are essential preventive measures. Considering our patient's background of rheumatoid arthritis and a history of long-term corticosteroid use, which can impair wound healing, such precautions are particularly important.[2,8]

In another study by Alexander et al,[9] the relationship between ACM-I and severe infections post-surgery is established. The results of this study align with our findings. In the case of our patient, despite appropriate treatment measures, she developed postoperative complications, including septic shock, which is consistent with the findings reported in the literature.[10] This comparison highlights that even when the best treatment practices are applied, ACM-I

can lead to severe and life-threatening complications. These findings emphasize the need for careful monitoring and continuous follow-up of patients after surgery and the importance of effective infection management.

Therefore, the results of previous studies have also shown that postoperative complications in patients with ACM-I are rare but potentially dangerous.[11] This case highlights the importance of heightened vigilance in postoperative care and the need for close monitoring of patients' status, especially in cases of CSF leakage and early signs of infection.[12,13] Additionally, there is a need for further research to explore more effective strategies for preventing and managing these complications.

In this case, the approach taken to manage and treat ACM-I and septic shock post-surgery was significant in several respects. First, the accurate and early diagnosis of Chiari malformation using appropriate imaging techniques was a strength in managing this case, which led to the correct decision for posterior fossa decompression surgery. Additionally, broad-spectrum antibiotics in managing postoperative infection, although they ultimately could not prevent the fatal septic shock, demonstrated the correct approach to infection control. Limitations of this case include the failure to prevent CSF leakage and the lack of timely infection control, both of which contributed to the patient's deteriorating condition.

The conclusion of this evidence-based study is derived from the patient's condition and their response to the treatments provided. The decision for decompressive surgery was made due to the severity of the patient's symptoms and confirmation of the diagnosis through imaging. Although appropriate therapeutic measures were implemented, CSF leakage and subsequent infection led to septic shock and, ultimately, the patient's death. These results indicate that even with proper management, the prognosis for patients with ACM-I can be challenging.

This case emphasizes the importance of precise management and continuous monitoring of patients after surgery for ACM-I. Previous studies have recommended thorough clinical examinations in the postoperative period to diagnose the most common complications, including aseptic meningitis, CSF leakage, infections, and hydrocephalus.[14,15] Therefore, specialized care to prevent infections and manage postoperative complications is essential. This study also highlights that awareness of the potential for septic shock and its early prediction can play a significant role in reducing mortality associated with this complication. As a result, more precise planning for preventing and managing postoperative complications can lead to better therapeutic outcomes.

Abbreviations

ACM-I, Arnold-Chiari Malformation Type I; CSF, Cerebrospinal fluid; ICU, Intensive Care Unit; CBC, Complete Blood Count; WBC, White Blood Count; RBC, Red Blood Cell; LDH, Lactate Dehydrogenase; B/C, Blood Culture Test; U/C, Urine Culture; CRP, C-reactive protein.

Data Sharing Statement

The data supporting this study's findings are available from the corresponding author upon reasonable request.

Ethics Approval

This study was conducted under the principles outlined in the Declaration of Helsinki. Ethical approval was obtained from the Ethics Committee of Tabriz University of Medical Sciences, with approval number (IR.TBZMED.REC.1402.840). According to institutional policies, this approval covers the entire research process, including the preparation and publication of case reports. Therefore, no additional institutional approval was required for publication.

Consent for Publication

The present study was conducted in one of the educational and medical centers in Iran. In these centers, during hospitalization, informed consent is obtained from all patients for the participation of students from different disciplines in the treatment processes of these patients and the use of their clinical records for education and research. In this study, written informed consent was obtained from the patient and his family for the use of their personal and clinical information in this case report, and it was ensured that confidentiality and anonymity were maintained. Since the patient

unfortunately died during hospitalization, due to the importance of the issue, informed consent was obtained again from the patient's family for its preparation, reporting, and possible publication.

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare that they have no competing interests in this work.

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