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Case Report

Management of Posttraumatic Cerebrospinal Fluid Leak with Secondary Diffuse Pneumocephalus Complicated by Meningitis and Communicating Hydrocephalus – Illustrative Case Report

Sintija Strautmane [1, 2, 3, †], Arturs Balodis [4, 5, †, *], Klavs Smidrovskis [1, 5, 6, †], Kaspars Auslands [3, 7, †]

1. Faculty of Residency, Riga Stradins University, Riga, Latvia; E-Mails: sintijasstrautmane@gmail.com; smidrovskisk@gmail.com
2. Department of Neurology, Pauls Stradins Clinical University Hospital, Riga, Latvia
3. Department of Neurology and Neurosurgery, Riga Stradins University, Riga, Latvia; E-Mail: kaspars.auslands@rsu.lv
4. Department of Radiology, Riga Stradins University, Riga, Latvia; E-Mail: arturs.balodis7@gmail.com
5. Institute of Diagnostic Radiology, Pauls Stradins Clinical University Hospital, Riga, Latvia
6. Department of Pathology, Riga Stradins University, Riga, Latvia
7. Department of Neurosurgery, Riga East Clinical University Hospital, Riga, Latvia

† These authors contributed equally to this work.

* **Correspondence:** Arturs Balodis; E-Mail: arturs.balodis7@gmail.com

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Abstract

Rhinorrhea is a possible complication after different types of traumatic brain injury or neurosurgical procedures, such as skull base operations. Pneumocephalus is a rarely noted complication after severe traumatic brain injury, and it may be accompanied by meningitis and ventriculitis, especially when treatment has been delayed. Treatment of these entities includes conservative and surgical approaches. Pneumocephalus may result in neurologic

disturbances threatening a life. Therefore, active patient management with a multidisciplinary team is required to prevent poor outcomes. In the literature, limited cases of rhinorrhea and pneumocephalus are available, including in our country. Pneumocephalus may also occur spontaneously. In 2015, Pishbin et al. identified 10 cases of spontaneous pneumocephalus. The precise incidence of diffuse pneumocephalus after traumatic brain injury is unknown, reported as <1% of cases with rare complications. In this case, a 41-year-old male patient presented at a tertiary university hospital with the chief complaint of headache. A month prior to admission, the patient was discharged from another hospital with multiple skull and facial fractures, pneumocephalus, and traumatic subarachnoid hemorrhage in the left frontal lobe due to fights (Le Fort III). During the second hospitalization, the patient's clinical status deteriorated. A repeated brain CT demonstrated diffuse pneumocephalus. Rhinorrhea was still present. The external lumbal drainage procedure was performed without stopping the cerebrospinal fluid leak. In children, pediatric inferior turbinate hypertrophy is a frequent cause of nasal breathing difficulties. In this case, no such hypertrophy was observed. It should be considered a nasal obstructive disease not necessarily related to adult entities, frequently associated with other nasal or craniofacial disorders. Early diagnosis and endoscopic management of rhinorrhea, nasal obstruction, and associated complications is vital, as delays can lead to life-threatening issues like hydrocephalus/meningitis. Eventually, the patient developed meningitis and acute communicating hydrocephalus. Right ventriculostomy with a programmable ventriculoperitoneal shunt placement was done (pressure 110 cm H_2O), stopping the rhinorrhea. This is an extremely rare case where a patient, after cerebrospinal fluid (CSF) leakage, develops severe complications, including pneumocephalus, meningitis, ventriculitis, and acute communicating hydrocephalus. In the literature, we did not come across case reports presenting all the complications as in this case. This case report will raise knowledge and awareness of such entities, adding to the rare, similar cases reported so far.

Keywords
Rhinorrhea; pneumocephalus; meningitis; communicating hydrocephalus; external lumbal drainage; neurosurgery; ventriculostomy; ventriculoperitoneal shunt

1. Case Presentation

A 41-year-old male was admitted to a tertiary university hospital with multiple skull fractures after a fight (Le Fort III) in October 2023. During this period, the patient exhibited somnolence and responded to medical inquiries that were incongruous and illogical. Brain computed tomography (CT) revealed multiple skull and facial fractures, including skull base, right temporal bone fracture, dislocated right mandibular angle fracture, as well as traumatic subarachnoid hemorrhage in the left frontal lobe and traumatic brain injury (see Figure 1, Figure 2); blood tests revealed ethanol use. The patient was hospitalized after being consulted by a neurosurgeon and an oral-maxillofacial surgeon with recommendations for conservative treatment. Nevertheless, facial bone osteosynthesis with mini and microplates was performed under general and local anesthesia, and the early postoperative period occurred without complications.

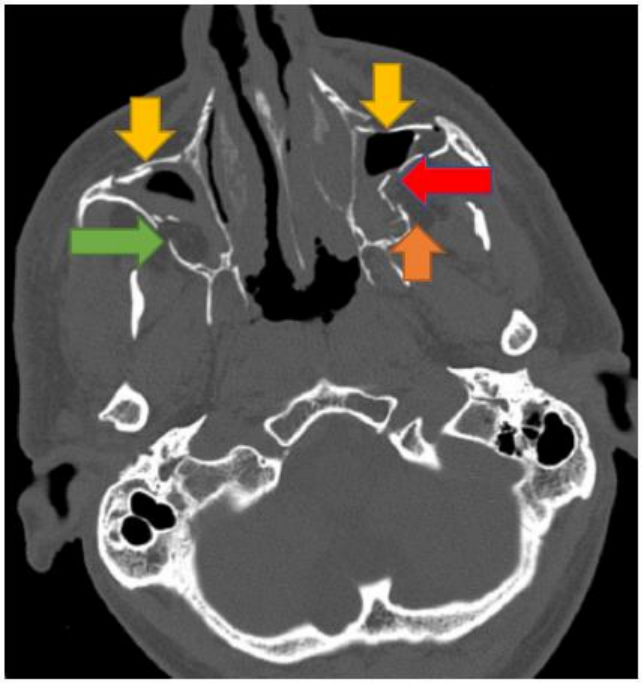

Figure 1 First patient non-enhanced brain computed tomography (NECT) after trauma revealing multiple skull and facial fractures; arrows indicating maxillary cavity fractures (bone window), October 2023.

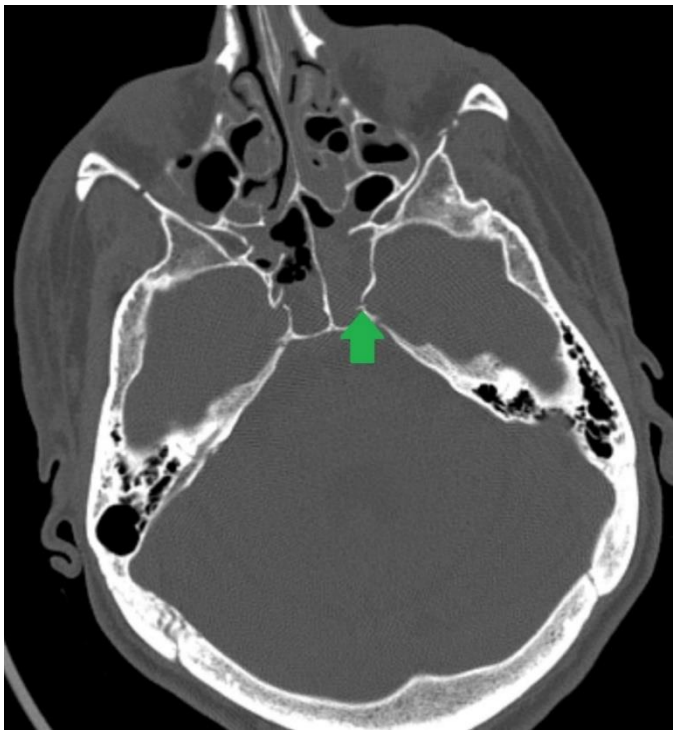

Figure 2 First NECT after trauma demonstrating bone fracture in the lateral wall of the sphenoid cavity with a little fragment and a little impact on the left internal carotid artery intracranially, October 2023.

After the procedure, the patient developed vertigo and progressive pulsating headache. A neurologist saw the patient; further recommendations were given. One day later, the patient

developed rhinorrhea from the left side of the nose, prompting the prescription of antibacterial therapy by an ear-nose-throat (ENT) specialist. A repeated brain CT was performed 4 days later, demonstrating complication – air diffusely within the cerebral cavity (pneumocephalus), most likely origin from a sphenoidal sinus (see Figure 3, Figure 4). The CT revealed blood in the prepontine cistern and the sphenoid sinus cavity (Figure 5). Additionally, a sphenoid cavity fracture was observed in the CT angiography; however, no evidence of a local posttraumatic lesion of the left internal carotid artery was noted (refer to Figure 6). A neurosurgeon and ENT specialist repeatedly consulted the patient to consider endoscopic closure of rhinorrhea. Considering the patient was clinically stable, further recommendations for conservative therapy were given with a planned visit to an outpatient clinic a month after this hospitalization to see if the rhinorrhea was still present.

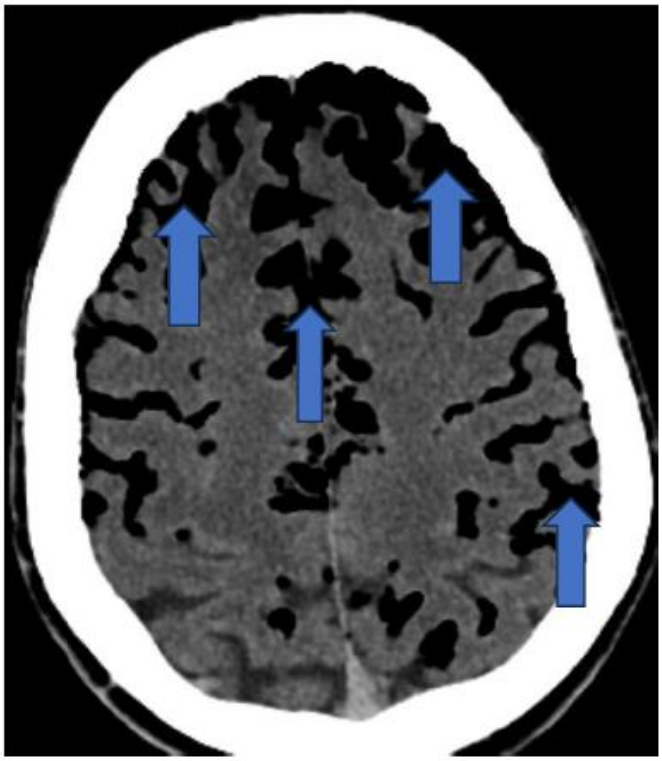

Figure 3 Pneumocephalus, seen in the repeated brain NECT 4 days after trauma (brain window; blue arrows), October 2023, 4 days after the previous NECT.

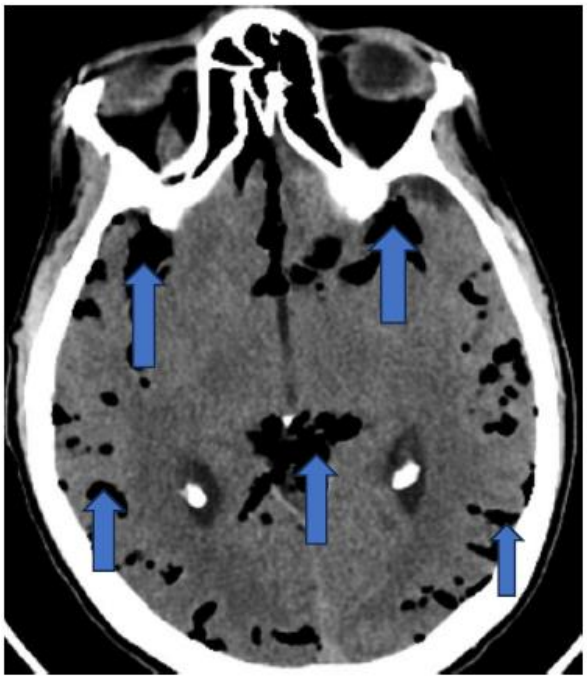

Figure 4 Pneumocephalus, seen in the repeated brain NECT 4 days after trauma (brain window; blue arrows), October 2023, 4 days after the previous NECT.

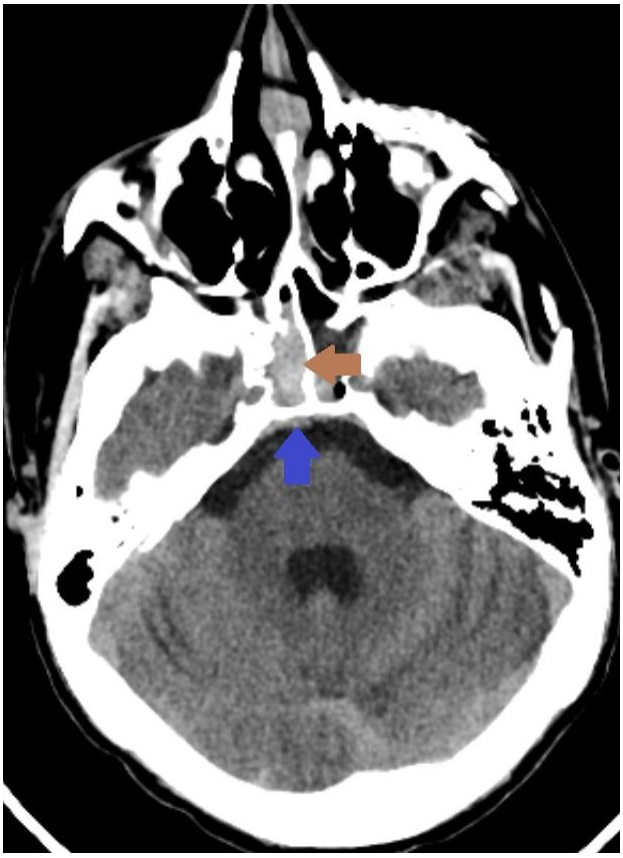

Figure 5 Blood in the sphenoid cavity (brown arrow) and in the prepontine cistern (blue arrow) seen in the same NECT 4 days after trauma, October 2023, 4 days after the previous NECT.

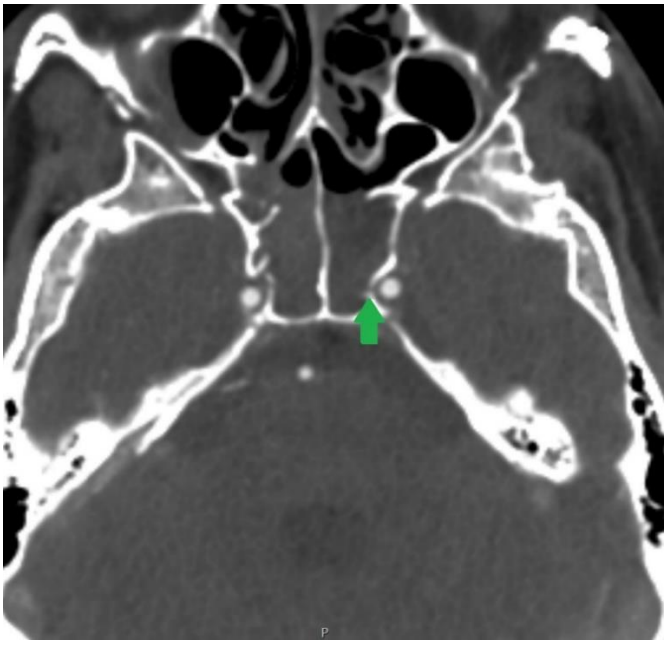

Figure 6 CT angiography, performed 4 days after trauma, revealing sphenoidal sinus wall fractures. No data of left internal carotid artery local posttraumatic lesion, October 2023, 4 days after the previous NECT.

A month later, in early December 2023, the patient was admitted to another tertiary university hospital with the main complaints of progressive headache and persistent rhinorrhea. A neurologist and a neurosurgeon consulted the patient. At the admission, the overall patient's condition was moderately severe, hemodynamically stable, and subfebrile temperature. The Glasgow coma scale was 15, and the patient's face was symmetrical. The pupils were centrally located and symmetrical, with a persistent reaction to light. In the patient's limbs, no paresis was observed. Coordination tasks in both arms and legs were performed precisely. Symmetrical deep tendon reflexes were observed, with no demonstration of pathological reflexes. The superficial and deep sensation was intact. The patient was further consulted by a neurosurgeon and hospitalized in the Neurosurgery ward, and antibacterial therapy was initiated.

During the hospitalization, the cerebrospinal fluid rhinorrhea was clear and still present. Six days after re-admission, external lumbal drainage was performed. The following spinal tap revealed pleocytosis (972). After the procedure, the patient exhibited recurrent episodes of headache and neck pain, along with noted positive meningeal signs. Rhinorrhea was still present after the external lumbal drainage procedure. A few days later, two weeks after being re-admitted to the hospital, the patient's clinical status deteriorated; the patient became markedly drowsy, and verbal communication with the patient became very challenging. A repeat brain computed tomography revealed diffuse pneumocephalus and abnormalities in cerebrospinal fluid circulation, displaying signs of transtentorial herniation and brain edema. Progressive widening of cerebral ventricles was noted, as well as periventricular edema, most likely acute communicating hydrocephalus due to meningitis (see Figure 7, Figure 8).

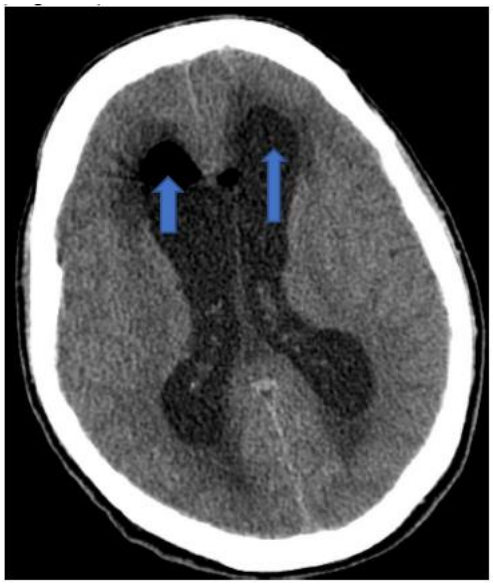

Figure 7 Repeated NECT 20 days after re-admission to the hospital demonstrating widening of cerebral ventricles (blue arrows), periventricular edema, most likely, acute communicating hydrocephalus due to meningitis, diffuse brain edema, cerebral sulci not visible, December 2023.

Figure 8 The same date brain NECT 20 days after re-admission to the hospital at the pons level revealing air at the prepontine cistern and air in the lateral ventricle temporal horns, December 2023.

A neurosurgical approach was indicated, leading to the patient undergoing right ventriculostomy under general anesthesia with an antibacterial drain on the same day after the results of repeated NECT scans (see Figure 7, Figure 8). The patient's clinical status stabilized, and the rhinorrhea gradually ceased. Another surgical intervention was indicated. Ventriculoperitoneal shunting

operation with a programmable (antibacterial) shunt was performed 12 days after ventriculostomy, pressure 110 cm H_2O, and the early postoperative period occurred without complications. The patient was gradually activated. Another brain computed tomography was repeated 8 days after the neurosurgical approach, demonstrating brain ventricles around the same size as before the surgery with a slight reduction. Uneven frontal edema in the periventricular regions was observed, along with diffuse brain edema without the danger of herniation (see Figure 9, Figure 10).

Figure 9 Patient brain NECT 8 days after the previous NECT, after right ventriculostomy; ventricles decreased in size, and less air is seen in them; less diffuse brain edema (red arrow), end of December 2023.

Figure 10 Patient brain NECT 8 days after acute communicating hydrocephalus and after right ventriculostomy (brain window): in dynamics, less edema diffuse brain edema, less widening of the temporal horns, no herniation, end of December 2023.

2. Results

This clinical case report pertains to a 41-year-old patient with a positive medical history involving multiple skull and facial fractures (Le Fort III), a traumatic subarachnoid hemorrhage in the left frontal lobe, and traumatic brain injury resulting from altercations in autumn 2023; concurrent use of ethanol was noted. During this period, the patient received initial conservative treatment while hospitalized. However, subsequent consultation with an oral-maxillofacial surgeon resulted in the performance of facial bone osteosynthesis using mini and microplates under general and local anesthesia. Repeated brain computed tomography revealed pneumocephalus, and the rhinorrhea was still present, but the patient was discharged from the hospital in a good overall health condition. A month after this episode, the patient was re-admitted to another hospital due to progressive headache, subfebrile temperature, and persistent rhinorrhea. External lumbal drainage was initiated. Following the procedure, the patient's condition worsened, displaying positive meningeal signs. A cerebrospinal fluid analysis revealed pleocytosis (972 cells), suggesting acute communicating hydrocephalus likely secondary to meningitis and ventriculitis. Transtentorial herniation, brain, and periventricular edema were also observed. In December 2023, a right ventriculostomy was conducted with the placement of an antibacterial drain. Subsequently, a ventriculoperitoneal shunting operation with a programmable shunt was performed with a pressure setting of 110 cm H_2O. Eight days after the neurosurgical treatment, the NECT brain showed the same ventricular enlargement with a slight reduction, uneven edema in periventricular regions, and diffuse edema without signs of herniation. The patient gradually improved. Early diagnosis and management of rhinorrhea is vital as delays can lead to life-threatening issues like hydrocephalus/meningitis. This is the first case report in our country about a patient demonstrating rare complications after traumatic brain injury, including rhinorrhea, pneumocephalus, acute communicating hydrocephalus, meningitis, and ventriculitis, with multiple illustrative materials.

The study follows the Code of Ethics of the World Medical Association (Helsinki Declaration). The authors obtained informed consent from the patient to publish a case report. The patient's anonymity is preserved.

3. Discussion

In a situation where cerebrospinal fluid (CSF) leaks extracranially into the paranasal sinuses and nasal cavity, finally exiting via the anterior nares, it is called CSF rhinorrhea [1-4]. According to the available literature data, it is a rare complication after severe traumatic brain injury, especially skull base fracture or iatrogenic causes, such as neurosurgical procedures, for example, transsphenoidal pituitary surgeries, complex operations at the skull base and otolaryngology procedures such as septoplasty and endoscopic surgeries [5, 6]. Nevertheless, the most noted cause of CSF rhinorrhea is closed-head trauma with an anterior base of skull fractures [7].

Rhinorrhea may also be caused by multiple congenital causes, for instance, meningocele, encephalocele, and lateral craniopharyngeal canal. It may also occur spontaneously due to idiopathic intracranial hypertension with medial sphenoid meningocele formation or in malignant nasopharyngeal and skull base tumors invading or involving the skull base [1, 8, 9].

Most frequently, patients with rhinorrhea may present intermittent discharge of clear fluid from the nose, often changing with the patient’s position [10-13]. The presentation typically occurs early, with most cases manifesting within the first 48 hours after the trauma or neurosurgical intervention

[11, 12]. It is possible to present later, but rhinorrhea beyond a year is rare [14]. Later presentations are associated with higher complication rates, including meningitis, which can lead to significant morbidity, as in the case demonstrated, as well as mortality [14]. Other complications of traumatic CSF leaks include recurrent, progressive headache and pneumocephalus, as well as neurological deficits, as in this case report.

In children, pediatric inferior turbinate hypertrophy is a frequent cause of nasal breathing difficulties [15]. In this case, no such hypertrophy was observed. It should be considered a nasal obstructive disease not necessarily related to adult entities, frequently associated with other nasal or craniofacial disorders. Early diagnosis and endoscopic management of rhinorrhea, nasal obstruction, and associated complications is vital, as delays can lead to life-threatening issues like hydrocephalus/meningitis [15].

Pneumocephalus, also known as pneumatocele or intracranial aerocele, refers to an air collection in the skull and brain parenchyma [16]. This condition should not be present under normal conditions. In the literature, 20% to 30% of patients with traumatic cerebrospinal fluid fistula develop pneumocephalus, and it can also occur in patients who do not have CSF rhinorrhea [16, 17]. According to the available literature data, pneumocephalus is found in 3.9% to 9.7% of patients with traumatic brain injuries, and it has also been reported to occur in 100% of cases following craniotomy [16-18]. Pishbin et al. identified 10 cases of spontaneous pneumocephalus [19]. The precise incidence of diffuse pneumocephalus after traumatic brain injury is unknown, reported as <1% of cases with rare complications [20-22].

Pneumocephalus may result in neurologic disturbances, such as headache, delirium, seizures, agitation, deep tendon reflex abnormalities, altered mental status, and frontal lobe syndrome – nonspecific symptoms. Therefore, the diagnostics of this entity is challenging [22-26]. In severe cases, it may threaten life. Therefore, active patient management with a multidisciplinary team is required to prevent poor outcomes [22, 26]. Pneumocephalus is not a common condition, but it is vital to be aware of this entity and its management to prevent serious complications. The pathogenesis of pneumocephalus has not been fully understood. However, it is assumed to be caused by discontinuous exchange of air and CSF when the intracranial pressure is lower than the atmospheric pressure [23]. The first treatment of choice in pneumocephalus includes antibacterial therapy, and it was the first treatment of choice in this clinical case report, but most patients require urgent neurosurgical treatment [22-26].

In this illustrative case report (see timeline in Figures above), a 41-year-old male patient with multiple skull and facial fractures, a subarachnoid hemorrhage in the left frontal lobe, and traumatic brain injury due to fights presented with persistent rhinorrhea and diffuse pneumocephalus upon discharge from the hospital. In this case, pneumocephalus presented with nonspecific symptoms, making the diagnosis challenging. One month after this episode, the patient was re-admitted to another hospital with acute communicating hydrocephalus due to meningitis and ventriculitis and received antibacterial treatment, as well as surgical treatment, including facial bone osteosynthesis with mini and microplates. Five days after re-admission, the patient underwent an external lumbal drainage procedure. Two weeks after being hospitalized, the patient deteriorated, repeated NECT demonstrated communicating hydrocephalus due to meningitis and diffuse brain edema, air in the prepontine cistern; therefore, on the same day, right ventriculostomy with a following ventriculoperitoneal shunting operation with a programmable (antibacterial) shunt placement was done later in the winter. This is an extremely rare case where a patient, after traumatic brain injury,

developed multiple severe complications, including rhinorrhea, pneumocephalus, acute communicating hydrocephalus, meningitis, and ventriculitis. Limitations of this case report include a lack of ability to generalize, no possibility of establishing a cause-effect relationship, and a lack of a comparison group.

The patient's overall clinical status stabilized, and the patient was gradually activated. This case report will raise knowledge and awareness of such entities, adding to the rare, similar cases reported so far.

4. Conclusions

Rhinorrhea is a rare, common complication after severe traumatic brain injury or iatrogenic causes, such as neurosurgical interventions, as well as non-traumatic etiologies, including idiopathic intracranial hypertension and tumors invading or involving the skull base. Pneumocephalus, on the other hand, is a condition noted seldom, but it may result in many neurological disturbances and threaten a life. Therefore, it requires knowledge and active surgical treatment to prevent serious complications and poor outcomes for the patient. In the literature, limited cases of rhinorrhea and pneumocephalus are available worldwide. There are extremely few cases reported with so many complications as in this case report. Therefore, this is one of the reasons why this case is so unique, providing a lot of clinical and radiological information to the readers. Key learning points of this case include awareness of non-specific symptoms of pneumocephalus resulting in its diagnostic challenge, as well as delays that can lead to life-threatening issues like hydrocephalus/meningitis. Therefore, knowledge of these conditions is vital. This case report will raise knowledge and awareness of such entities, adding to the rare, similar cases reported so far. Further investigations of rhinorrhea and pneumocephalus are warranted to understand their pathogenetic mechanisms and provide the patient with the best possible treatment.

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Author Contributions

All authors contributed to the article's conception and design. Material preparation, data collection and analysis were performed by SS, AB, KA, and KS. The first draft of the manuscript was written by SS. It was reviewed and corrected by SS and AB. All authors read and approved the final manuscript.

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Competing Interests

The authors have declared that no competing interests exist.

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