

## A Rare Case of Spontaneous Pneumocephalus as a Complication of Nontraumatic Nasal Liquorrhea

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### Abstract

Pneumocephalus is the ingress of air into the intracranial cavity. Pneumocephalus is associated with several etiological factors, such as head injuries, surgical interventions, infections and neoplasms. With spontaneous nasal liquorrhea, pneumocephalus is extremely rare, since a defect must be large in order to cause it. Clinical implications of pneumocephalus depend on location and volume of air in the cranial cavity. The most common and described symptoms are headache, "splashing sound", rhinorrhea and otorrhea, meningism, dysfunction of cerebrospinal nerves, hemiparesis, optic disc edema, epileptic seizures, collaptoid states, psychiatric symptoms. For treatment of pneumocephalus, both conservative and surgical methods are used. The choice of tactics depends on type, etiology and volume of the air that has entered the cavity of the skull.

In this article, we describe the case of effective treatment of spontaneous nasal liquorrhea, complicated by pneumocephalus and meningitis.

**Keywords:** Pneumocephalus; Cerebrospinal fluid leak; Fluorescein; Psychopathological symptomatology in pneumocerephaly skull base surgery

### Introduction

The aim of the study was to describe successful treatment strategy for nontraumatic nasal liquorrhea complicated with pneumocephalus. Spontaneous nasal liquorrhea is a rare condition which can lead to serious complications like meningitis and pneumocephalus. If this condition stays undiagnosed, it will be fatal for the patient. There has been observed a lack of information about this condition and no concerted treatment strategy up to date [1]. We found nine clinical cases with the same condition. Consequently, conventional algorithm should be developed generalized from recorded cases.

### Case Report

A 57-year-old patient was admitted to National Scientific and Practical Center of Neurosurgery named after academician NN Burdenko in the department of neurotrauma in a serious condition.

### Life history

Childhood infections, for 40 years hypertension of the 3<sup>rd</sup> degree 3 d. risk 4. Craniocerebral injuries and allergic reactions are denied.

### Medical history

One year ago, the patient began to notice the flow of clear fluid from the left nasal passage, which periodically spontaneously ceased, then again recurred. Two months before admission she noticed headaches, fever, nausea, vomiting.

Hospitalized at the place of residence. According to the SCT of the brain-pronounced pneumocephalus. The operation "endonasal closure of the liquor fistula with the adhesive composition Onyx" was performed. A course of antibiotic therapy was conducted. Eight days after the operation there was a worsening of the condition in the form of a decrease in the level of wakefulness, in connection with which ventriculopuncture was performed without effect. Immediately hospitalized in Federal State Autonomous Institution, National Scientific and Practical Center of Neurosurgery named after academician NN Burdenko in the department of neurotrauma [2,3].

### Objective data on admission

Serious condition, level of consciousness: stunning, drowsiness, lethargy. The Karnofsky index is 50%. Heart sounds are clear, rhythmical. Blood pressure 140/80 mm Hg. Does not control stool and diuresis. Simple instructions are carried out. There is stiff neck. The volume of movements in the limbs is not limited [4]. Reflexes of average liveliness on both sides. Can not stand. There is a sutured wound in the right frontal region after ventriculopuncture.

### ENT-examination

There are no signs of nasal liquorrhea.

The SCT of the brain shows destructive changes in the posterior wall of the main sinus, with the presence of the exudative component in the left parts of the main sinus, the latticed labyrinth, the posterior parts of the left maxillary sinus. In the ventricular system, basal cisterns, anterior sections of the frontal lobes, the accumulation of air is determined. In the right frontal region, there is a bone defect, most likely of postoperative genesis. The ventricular system is not enlarged, except for the anterior horns of the lateral ventricles, in which there is an accumulation of air. Convex subarachnoid spaces are not expanded [5,6]. The median structures are not biased. Basal tanks are traced (Figures 1-3). In the blood analysis upon admission-leukocytosis 10.31 ESR 17.

### Clinical diagnosis

Complex skull base defect. Spontaneous relapsing nasal liquorrhea. Massive pneumocephalus.

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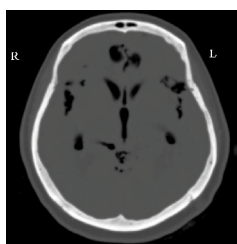
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**Figure 1:** CT scan of the brain (frontal projection).



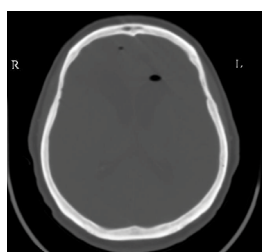
**Figure 2:** CT scan of the brain (axial projection).



**Figure 3:** CT scan of the brain (sagittal projection). The arrow indicates a bone loss.



**Figure 4:** Defect in the sphenoidal sinus.



**Figure 5:** CT scan- two days after surgery (axial projection).

## Course of treatment

Under general anesthesia, the operation “Endoscopic endonasal plastic of a complex skull base defect in the region of the sphenoid sinus on the left under the control of the navigation system” was performed.

Lumbar puncture was performed during the operation. There was no cerebrospinal fluid, which was regarded as a “dry lumbar puncture”, thereby subdural injection of 1.0 ml of 5% of Fluorescein sodium was done. The consent of the patient’s relatives was received, there were no side effects during the injection and operation. In the region of the posterior wall of the sphenoid sinus, a pocket with two compartments was defined. In the left one, after removal of the mucosa, a bone defect 1 mm in diameter was exposed, from which the colored liquor was emitted with a pulsation (Figures 4).

When checking the navigation system, the defect coincided with the CT data. Plasticity was carried out by fragments of autofascia and auto-fat taken from the middle third of the left thigh, fixed by fibrin-thrombin sealant “Evicel”.

In the postoperative year in the 1<sup>st</sup> day the condition remained critical. The level of consciousness is stunning. Rigidity of the occipital muscles. Kernig’s symptom is doubtful. The temperature is 38.5. In a general blood test, leukocytosis is 18.40, ESR 14. General analysis of cerebrospinal fluid: cloudy, protein 1.07%, cytosis 487/3, lymphocytes 36%, neutrophils 58%. In the biochemical analysis of blood, C-reactive protein 106.4. The patient underwent massive antibacterial, anti-inflammatory, hyposensitizing and detoxification therapy. Vancomycin 1 g twice a day and Doriprex 1 g three times a day for 2 weeks were prescribed.

According to the CT data, air disappearance in basal cisterns with a significant decrease in the ventricular system and frontal area was observed on the 2<sup>nd</sup> day after the operation. The lateral ventricles are dilated, mainly in the anterior parts (Figure 5).

## Mental specialist examination

According to the medical staff and relatives, the patient had episodes of anxiety, she took out a cubital catheter. She lies in bed, reacts to the spoken speech, turns the head towards the interlocutor. Contact with the patient is difficult. After repeated requests, one can get monosyllabic answers to some questions. During the conversation, the patient periodically begins to laugh, can not explain a reason for laughing. Can not remember the current year, month, date. She considers herself being in the house of culture. Conclusion: confusion on the background of regress of spontaneity.

Against the backdrop of ongoing therapy for 16 days, the patient’s condition improved significantly: T body normalized, somatically and neurologically stable. Meningeal signs are absent. She walks with support. With ENT-examination there are no signs of nasal liquorrhea. Mental specialist examination: the patient is in a clear mind, disorientation is preserved. The instructions are clearly implemented. Clinical performance is normal.

Dynamics of cerebrospinal fluid parameters is presented in Table 1.

Before discharge the CT shows lack of air in the basal cisterns, ventricular system and frontal region. Basal cisterns are not deformed, subarachnoid spaces are not expanded.

The patient is discharged to the polyclinic at the place of residence under the care of a physician.

## Discussion

In this clinical example, we observed a rare case of spontaneous pneumocephalus. The term “spontaneous pneumocephalus” is applied to a state where air is accumulated intracranially regardless of any cause: tumor, infection, inflammation, surgery, or trauma [7] In this patient,

Postoperative period	Protein	Cytosis	Color	Transparency	Deposit	Lymphocyte	N. Number	Glocose	Lactate
The 2 <sup>nd</sup> day	107%	484/3	Pink	Cloudy	white-red	36%	58%	1.2	5.1
The 5 <sup>th</sup> day	136%	428/3	Sl. yellow	Full	--	82%	18%	1.9	3.8
The 10 <sup>th</sup> day	168%	71/3	Sl. Yellow	W.c.	barely red	63	7	2.0	3.1
The 15 <sup>th</sup> day	059%	17/3	Barely yellow	Full	--	16	--	2.1	3.0

**Table 1:** General analysis of cerebrospinal fluid in dynamics and biochemical analysis of cerebrospinal fluid in dynamics.

Authors	Age	Gender	Defect localization	Predisposing causes	Symptoms	Mental disorders	Hyperpneumatization of sinuses	Concomitant conditions	Treatment	Application of Fluorescein
Wakefield [8]	28	M	Sphenoidal sinus	Cough Sneezing	Headaches	Confused mental state	-	Allergic rhinitis	Surgical treatment	-
Mirone [9]	50	M	Ethmoid sinuses	Valsalva's manoeuvre	Loss of consciousness	MMSE 15 out of 3 <sup>0</sup> 1	-	Obsessive-compulsive disorder	Surgical treatment	-
Lee et al. [10]	31	M	Sinus of scull and sphenoidal sinus	-	Headaches, nausea	-	+	-	Conservative treatment	-
Park [11]	35	M	Sinus of scull	Nose blow	Headache	-	+	-	Conservative treatment	-
Shete [12]	61	F	Sphenoidal sinus	Barotrauma	Headache, nausea, vomiting	Disorientation, confused mental state	+	meningitis	Surgical treatment	+
Kim et al. [13]	62	F	-	-	Headaches	Disorientation, confused mental state	-	meningitis	Conservative treatment	-
Pishbin [14]	51	F	Sphenoidal sinus	Cough	Headaches, nausea	-	-	-	Conservative treatment	-
Baba [15]	51	F	Sphenoidal sinus	Sneezing	Headaches	-	+	Liquorrhea Idiopathic intracranial hypertension	Surgical treatment	+
случай	57	F	Sphenoidal sinus	-	Headache	Disorientation, confused mental state	+	Liquorrhea, meningitis	Surgical treatment	+

<sup>1</sup>The MMSE indicator is a scale for assessing mental status. F– Female, m– Male.

**Table 2:** Summary of cases with spontaneous pneumocephalus.

pneumocephalus developed against a background of spontaneous nasal liquorrhea. Most likely in the pathogenesis there was a mechanism of “inverted bottle” or “siphon effect”, when a prolonged profuse nasal liquorrhea in the cranial cavity created a negative pressure and through the existing defect the air came in an extracranial manner. The small size of the defect (1 mm) is relevant in this case, also the fact of cessation of increased production of cerebrospinal fluid in response to its chronic loss, as it usually happens in other patients with nasal liquorrhea.

Massiveness of pneumocephalus with increasing clinical symptoms has been developing in this patient for 2 months. The main symptoms are headache, nausea, vomiting, raising the temperature of the body with subsequent oppression of consciousness. Of interest is the dynamics of psychiatric symptoms in our patient. In the preoperative period, asponaneity, lethargy, drowsiness, blockade of voluntary activity, fulfillment of only simple instructions without other manifestations of contact, impossibility of independent walking was noticed. After the operation, asponaneity regressed and was replaced by episodes of anxiety, increased contact, it became possible to obtain monosyllabic answers to questions. However, against the background of these positive changes, disorientation in the surrounding area was revealed (so, when asked about the location, the patient responded that she was “in the house of culture”) and there was a violent laughter, the appearance of which may indicate the irritation of the subcortical structures that followed their oppression. After a week, the confusion regressed a little, the patient began to walk with support, but remained disoriented.

Thus, against the background of regression of pneumocephalus,

positive dynamics in the mental status in the form of regress of the oppressed consciousness with asponaneity, transition to the next stage of restoration of mental activity - the state of the disintegrated consciousness, was noted.

It is worth mentioning the role of endolyumbellation of fluorescein sodium 10% during the operation. It was possible not only to confirm the presence of liquorrhea, which, considering pneumocephalus, was known, but it was also correct to localize the site of the defect, despite the altered anatomy of the nasal cavity after the first operation and the individual features of the structure of the sphenoid sinus. However, it is worth noting that, since the cerebrospinal fluid pressure was low, the lumbar puncture was dry. In order to properly follow the procedure for the administration of fluorescein and to avoid undesirable side effects in such cases, one must have a lot of experience.

According to the world literature, spontaneous pneumocephalus is very rare. From 1996 to 2016 we found the description of 8 clinical cases of this pathology, the summary data of which are given in Table 2 [8-15].

From the data in the table it follows that most often the cause of spontaneous pneumocephalus is a bone defect in the sphenoid sinus, as in our patient. In this pathogenesis, the mechanism of the “air valve” predominates with an extracranial increase in pressure due to blowing, sneezing, barotrauma, and stress tests. In clinical symptoms, as in our case, headache, nausea, vomiting and psychiatric symptoms in the form of disorientation, confusion of mind prevail. The nasal liquorrhea is described in both our and also in another case, meningitis was

observed in two patients. In most cases (5 out of 9) surgical tactics with endoluminal administration of Fluorescein in three patients were used.

We found some limitations in description of previous clinical cases and tried to expand some information about this condition. Mental disorders are frequent enough among patients with meningitis and pneumocephalus, consequently dynamic psychological supervision should be performed. Only in our clinical case the changes in psychological status was described. We expanded description of pathophysiology of nontraumatic nasal liquorrhea complicated with spontaneous pneumocephalus in more details. Furthermore, we did not find detailed description of Fluorescein injection in patients with pneumocephalus in referenced articles.

We are going to continue work with these patients and develop algorithm of treatment such patients base on our and international experience.

## Conclusion

In the case of nasal liquorrhea in an anamnesis, if the patient's condition worsens in the form of development of cerebral and psychiatric symptoms, it is worth to remember of possible development of pneumocephalus, the early diagnosis of which will help to choose the correct therapeutic approach.

With the proven diagnosis of "spontaneous pneumocephalus" with the revealed skeletal defect of the base of the skull according to CT and nasal liquorrhea in anamnesis, surgical treatment is a method of choosing and preventing infectious complications.

Intralumbar injection of 10% sodium fluorescein may be useful for intraoperative defect localization in the case of pneumocephalus, even with the absence of nasal liquorrhea at the time of surgical treatment.

Pneumaticcephaly can be manifested by a change in the oppressed consciousness disintegrated, with the transition from a blockade of arbitrary activity to a rapidly regressing confusion of consciousness

## Disclaimer

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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