PNEUMOLABYRINTH AS A POSTOPERATIVE COMPLICATION OF STAPEDOPLASTY

Diab KhMA[1,2] ✉, Daikhes NA[1], Pashchinina OA[1], Zukhba AG[1], Kokhanyuk SV[1], Pirogova NE[2]

[1] National Medical Research Center for Otorhinolaryngology of the Federal Medical Biological Agency, Moscow, Russia

[2] Pirogov Russian National Research Medical University, Moscow, Russia

Pneumolabyrinth is characterized by the presence of air in the inner ear due to intrusion from the tympanic cavity. It is a rare complication of stapedoplasty. Currently, there is no clear algorithm for treatment of this complication. The paper reports two cases of pneumolabyrinth being the short-term and long-term postoperative complications. In the first case, the patient, who had undergone stapedoplasty in the left ear, suddenly developed rapid hearing loss and tinnitus in the left ear with dizziness three weeks after physical activity. Physical examination revealed no evidence of the tympanic membrane defect. Audiometry revealed left-sided IV degree of sensorineural hearing loss. Pneumolabyrinth was detected on the temporal bone CT scans. In the second case, vestibulocochlear symptoms developed three days after stapedoplasty in the right ear. Pure tone audiometry revealed right-sided IV degree of mixed hearing loss. CT scan of the temporal bone confirmed the diagnosis of pneumolabyrinth. In both cases the correct position of the stapedial prosthesis, "empty" vestibule and perilymphatic fistula were found during revision tympanotomy. The prosteses were removed during surgery, Dexamethasone solution was introduced into the vestibule; stapedoplasty with autocartilage on the perichondrium was performed. After surgery, vestibular symptoms disappeared, and hearing improved.

Keywords: pneumolabyrinth, stapedoplasty, perilymphatic fistula, vestibulocochlear symptoms, pneumatization, sensorineural hearing loss

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✉ **Correspondence should be addressed:** Khassan M. A. Diab
Volokolamskoe shosse, 30, str. 2, Moscow, 123182, Russia; hasandiab@mail.ru

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ПНЕВМОЛАБИРИНТ КАК ОСЛОЖНЕНИЕ ПОСЛЕ СТАПЕДОПЛАСТИКИ

Х. М. А. Диаб[1,2] ✉, Н. А. Дайхес[1], О. А. Пащинина[1], А. Г. Зухба[1], С. В. Коханюк[1], Н. Е. Пирогова[2]

[1] Национальный медицинский исследовательский центр оториноларингологии Федерального медико-биологического агентства, Москва, Россия

[2] Российский национальный исследовательский медицинский университет имени Н. И. Пирогова, Москва, Россия

Пневмолабиринт характеризуется наличием воздуха во внутреннем ухе вследствие его проникновения из барабанной полости. Он является редким осложнением стапедопластики. В настоящее время нет четкого алгоритма лечения этого состояния. В статье рассмотрены два случая пневмолабиринта, возникшего как раннее и позднее осложнения после операции. В первом случае у пациентки, перенесшей стапедопластику на левом ухе, через три недели после физической нагрузки внезапно развились резкое снижение слуха на левом ухе, шум в левом ухе и головокружение. При физикальном осмотре не было обнаружено никаких признаков дефекта барабанной перепонки. На аудиометрии диагностирована левосторонняя сенсоневральная тугоухость IV степени. По данным компьютерной томографии (КТ) височной кости выявлен пневмолабиринт. Во втором случае развитие вестибулокохлеарной симптоматики имело место спустя три дня после стапедопластики на правом ухе. На аудиометрии обнаружена правосторонняя смешанная тугоухость IV степени. Данные КТ височной кости подтвердили диагноз пневмолабиринта. В обоих случаях во время ревизионной тимпанотомии был обнаружен стапедиальный протез, установленный проксимальным концом в преддверие и дистальным концом закрепленный на длинном отростке наковальни, а также перилимфатический свищ. В ходе операции протез удалили, в преддверие ввели раствор дексаметазона, выполнили стапедопластику по методике «аутохрящ на надхрящницу». После операции вестибулярные симптомы исчезли, слух несколько улучшился.

Ключевые слова: пневмолабиринт, стапедопластика, перилимфатический свищ, вестибулокохлеарные симптомы, пневматизация, сенсоневральная тугоухость

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✉ **Для корреспонденции:** Хассан Мохамад Али Диаб
Волоколамское шоссе, д. 30, стр. 2, Москва, 123182; hasandiab@mail.ru

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Pneumolabyrinth is a rare condition characterized by substitution of perilymph that fills the labyrinth with air due to abnormal communication between the middle and inner ear. In other words, pneumolabyrinth is the presence of air in the inner ear that results from communication between the inner ear and middle ear that is filled with air. Clinical manifestations of the condition are associated with such vestibulocochlear disorders, as sensorineural hearing loss, “ringing” in the ears and/or head, ear fullness, as well as vertigo and nausea [1, 2]. The final diagnosis is established based on the data of high resolution computed tomography (HRCT) of the temporal bone by the presence of air in the inner ear.

The term “pneumolabyrinth” was introduced into the scientific literature by M.F. Mafee et al. [3]. Etiological factors that lead to the development of pneumolabyrinth conditionally can be divided into traumatic, iatrogenic and inflammatory factors. According to the statistical analysis of data obtained from electronic databases (PubMed, MEDLINE, EMBASE, Cochrane Library, Scopus), temporal bone fractures (31.8%), stapes surgery (18.2%), penetrating trauma (15.9%), and barotrauma

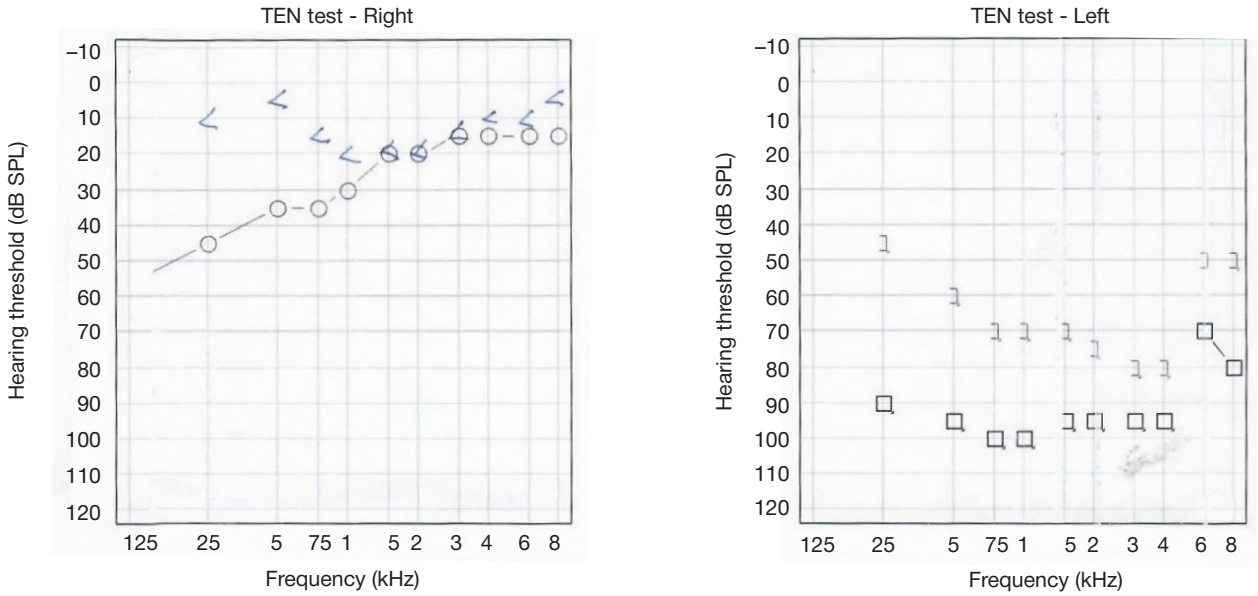

Fig. 1. PTA data of patient K., 32 years old, at admission (before revision tympanotomy)

(11.4%) are the most common causes of pneumolabyrinth. Based on the temporal bone CT scans, pneumolabyrinth, both as a solo disorder and in combination with other disorders, is most often found in the vestibule (95.3%), followed by cochlea (40.2%) and semicircular canals (23.4%) [4].

Long-term pneumolabyrinth is a rare complication of stapedectomy related to the prosthesis displacement or luxation. The condition should be considered in patients with vestibulocochlear symptoms, even many years after surgery. It should be noted that pneumolabyrinth that occurs within the first week after surgery suggests the presence of perilymphatic fistula. In the majority of cases reported in scientific literature, pneumolabyrinth occurred within weeks or months after stapes surgery. The article presents two cases of pneumolabyrinth.

Clinical cases

Pneumolabyrinth as a late complication of stapedoplasty

Female patient K. aged 32 was admitted to the Department of Ear and Skull Base Pathology of the National Medical Research Center for Otorhinolaryngology of FMBA with complaints of vertigo when changing body position and walking, tinnitus and hearing loss in her left ear. According to the history of the disease, the patient underwent left piston stapes surgery with the use of the SPL 03.44S prosthesis (Audio Technologies; Italy) (prosthesis length: 4.5 mm) on September 2, 2020; the perforation diameter was 0.6 mm. The patient was discharged from the hospital on September 7, 2020 with improved hearing in the operated ear. During the postoperative period (October 2020), she noted left-sided rapid hearing loss, tinnitus in her left ear, vertigo when changing body position and walking that developed after excercise. The patient received conservative therapy in the local clinic. She was referred to the outpatient clinic of the National Medical Research Center for Otorhinolaryngology of FMBA and then admitted to the Department of Otology and Skull Base Disorders due to persistent complaints.

Physical examination showed that both tympanic membranes were pearly grey, translucent, with clear margins, mobile, had no defects. Examination revealed no spontaneous nystagmus, a negative Rinne's test in the left ear, and sound lateralization to the right ear in the Weber's test. Pure-tone audiometry (PTA) showed right-sided mixed hearing loss, degree I, and left-sided sensorineural hearing loss, degree IV (Fig. 1).

The temporal bone CT scans revealed CT signs of bilateral fenestral otosclerosis, the condition after left stapedoplasty. Pneumatized antrum, mastoid cells and tympanic cavity were observed. Air was found in the cochlear duct and vestibule of the labyrinth on the left (Fig. 2).

The following clinical diagnosis was made based on the patient's complaints, disease history, PTA and the temporal bone CT scan: fenestral otosclerosis; condition after surgery (stapedoplasty) performed in the left ear on September 2, 2020; pneumolabyrinth; left-sided sensorineural hearing loss, degree IV.

The committee of physicians decided to perform left revision tympanotomy under local anesthesia (sol. Lidocaini 2% — 12 mL). Intrameatal approach through Rosen's incision performed with microsurgical technique was used under microscope. Meatotympanic flap was separated to the level of annulus fibrosus, tympanotomy was performed. Exploration of the tympanic cavity revealed the long process of the incus and malleus along with no stapes structures. The Audio Technologies SPL 03.44S prosthesis (4.5 mm) was installed with its proximal end in the vestibule and the distal end fixed on the long process of the incus. The oval window perilymphatic fistula was found. The prosthesis was removed.

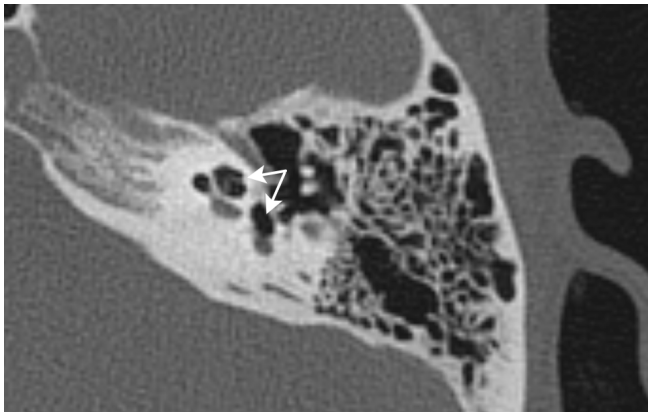

Fig. 2. CT scan at admission, left temporal bone of patient K. aged 32; the *arrows* indicate the presence of air in the cochlea and vestibule

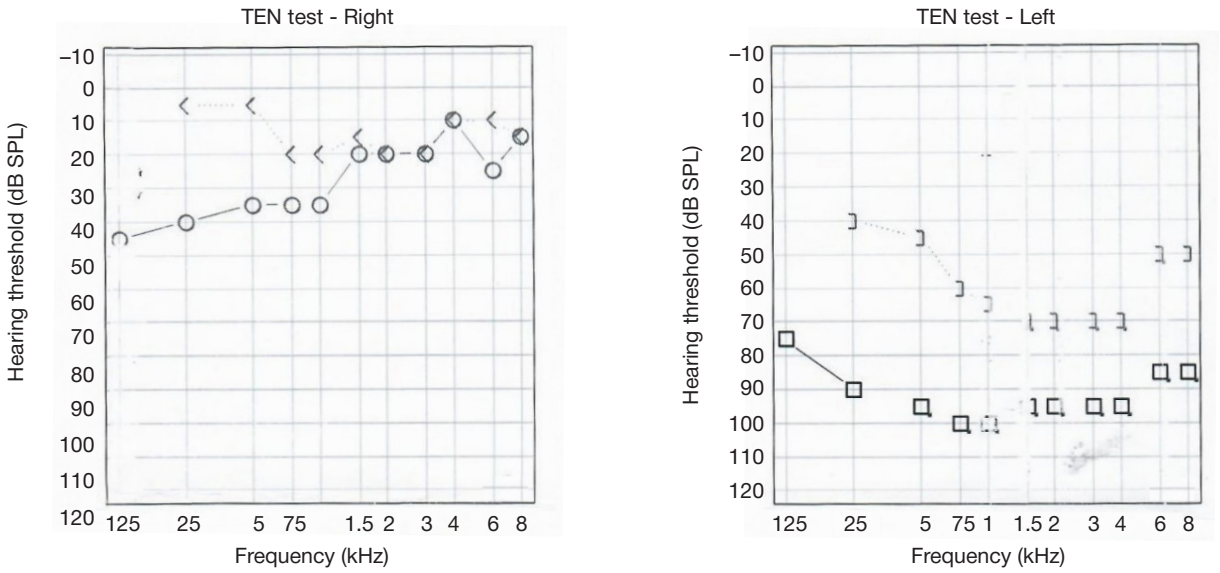

Fig. 3. PTA data of patient K., 32 years old, obtained in the short-term postoperative period

Autocartilage graft with perichondrium was taken from the auricle. Dexamethasone solution and normal saline were injected in the vestibule. A perichondrium flap was put on the area of vestibule at the level of window frame. An autocartilaginous columella with the length of 3.5 mm was installed with its proximal end on the above-mentioned peace of perichondrium and its distal end under the lenticular process of the incus. Meatotympanic flap was put back. Tamponade of the external auditory canal with the antibiotic-loaded (ceftriaxone) hemostatic sponge was peformed.

The patient was prescribed a course of conservative therapy: glucocorticoid drug (dexamethasone 24–20–16–8–4 mg + NaCl 0.9% 200.0 mL, intravenous infusion once a day); Betahistine (Betaserc) 24 mg three times daily; vitamin therapy (vitamins B1, B6 — 1.0 mL, intramuscular injection every other day); vascular therapy (actovegin 10.0 mL + NaCl 0.9% 10.0 mL, intravenous infusion); Choline Alfoscerate (Noocholin Rompharm) 4.0 mL (250 mg/mL) + NaCl 0.9% 100.0 mL, intravenous infusion.

The control PTA performed in the postoperative period revealed right-sided mixed hearing loss, degree I, and left-sided sensorineural hearing loss, degree IV (Fig. 3).

Another CT scan of temporal bones performed on December 4, 2020, revealed CT signs typical for the condition after left stapedoplasty (Fig. 4).

Four days after revision surgery, the patient noted no dizziness and improved hearing in her left ear. No signs of paresis of the mimic muscles or spontaneous nystagmus were revealed. The Weber's test showed sound lateralization to the left ear. It was decided to discharge the patient from the hospital for further ENT follow-up in the community clinic.

Pneumolabyrinth as a short-term complication of stapedoplasty

Female patient S. aged 37 was admitted to the Department of Ear and Skull Base Pathology of the National Medical Research Center for Otorhinolaryngology of FMBA with complaints of right-sided hearing loss, tinnitus in the right ear, and vertigo. Physical examination showed that both tympanic membranes were pearly grey, translucent, with clear margins, mobile, with no defects. There was no spontaneous nystagmus. The tuning fork tests showed a negative Rinne's test in the right ear, and sound lateralization to the right ear in the Weber's test. PTA revealed right-sided mixed hearing loss, degree III (Fig. 5).

The temporal bone CT scan performed on August 17, 2020 revealed CT signs of fenestral otosclerosis.

The following clinical diagnosis was made based on the patient's complaints, disease history, PTA and the temporal bone CT scan: fenestral and cochlear otosclerosis; right-sided mixed hearing loss, degree III.

On November 17, 2020, the patient underwent right piston stapedoplasty performed with the use of the Audio Technologies SPL 03.44S prosthesis (prosthesis length 4.5 mm) under local anesthesia. She noted hearing improvement in her right ear during the operation. Three days after surgery, the patient noted rapid hearing loss and tinnitus in her right ear, vertigo when changing body position, nausea, and high-amplitude horizontal right-jerk nystagmus that developed due to patient non-compliance (physical exercise).

The patient got the consultation of otoneurologist, and underwent PTA that revealed right-sided mixed hearing loss, degree IV, during the early postoperative period. The temporal bone CT scan was performed that revealed CT signs of bilateral fenestral otosclerosis, the condition after right stapedoplasty (air was found in the cochlear duct, vestibule of the labyrinth, and lateral semicircular canal) (Fig. 6).

The committee of physicians decided to perform revision tympanotomy. Intrameatal approach through Rosen's incision, performed through microsurgical technique, was used, that was controlled with the surgical microscope. Meatotympanic flap was separated to the level of annulus fibrosus, tympanotomy

Fig. 4. CT scan performed during the short-term postperative period, left temporal bone of patient K. aged 32: the *arrows* indicate the cochlea and vestibule filled with liquid content

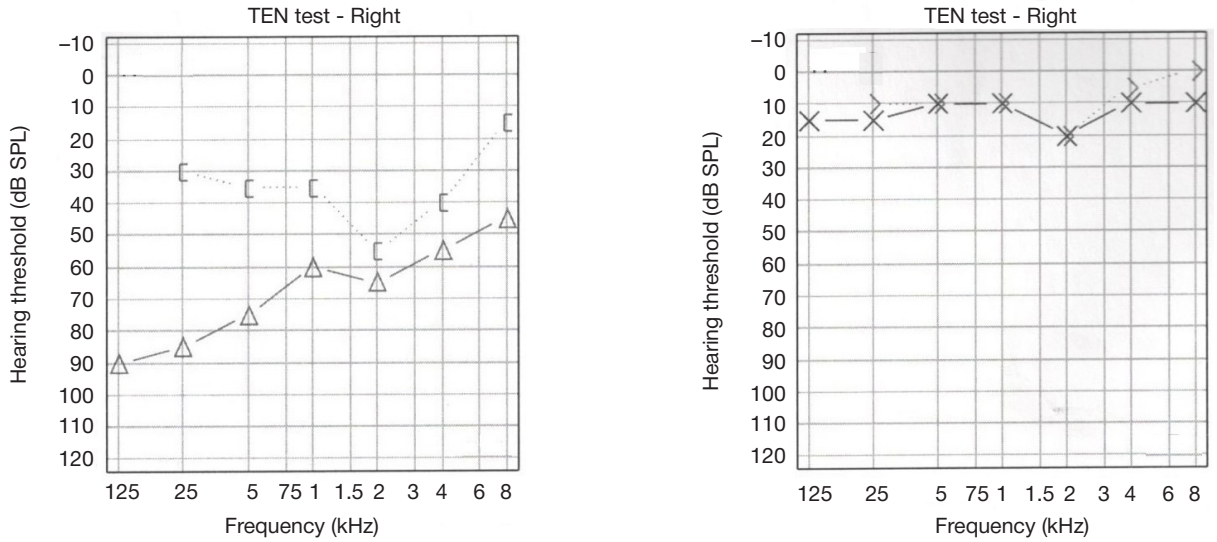

Fig. 5. PTA data of patient S., 37 years old, at admission

was performed under local anesthesia (sol. Lidocaini 2% — 12 mL). Exploration of the tympanic cavity revealed no stapes structures. The Audio Technologies SPL 03.44S prosthesis (4.5 mm) was installed with its proximal end in the vestibule and the distal end fixed on the long process of the incus. The prosthesis was removed. Autocartilage graft with perichondrium was collected from the auricle. Dexamethasone solution and normal saline were injected up to the level of the vestibule window frame. A piece of perichondrium was put on the area of the oval window niche. An autocartilaginous columella with the length of 3.5 mm was installed (Fig. 7). The meatotympanic flap was put back. Tamponade of the external auditory canal with the antibiotic-loaded (ceftriaxone) hemostatic sponge was peformed. A dry sterile auricular dressing was applied. No dizziness or spontaneous nystagmus was observed immediately after surgery.

The patient received a course of conservative therapy: dexamethasone 20–16–12–8–8–4 mg + NaCl 0.9% 200.0 mL, intravenous infusion; betahistine (Betaserc) 16 mg, three times daily; vitamin B12 250 mg, intramuscular injection once a day; choline alfoscerate (Noocholin Rompharm) 4.0 ml (250 mg) + NaCl 0.9% 100.0 mL, intravenous infusion; actovegin 10.0 mL + NaCl 0.9% 100.0 mL, intravenous infusion.

On 6 day after surgery, the patient noted improved hearing in her right ear, reduced dizziness, and reduced tinnitus in the right ear (Fig. 8).

Another CT scan of temporal bones revealed CT signs of fenestral otosclerosis, the condition after right stapedoplasty (Fig. 9).

It was decided to discharge the patient, who showed an obvious improvement, from the hospital for further ENT follow-up in the community clinic and subsequent consultations in the Center.

Clinical case discussion

Pneumolabyrinth is a rare condition associated with air trapped in the inner ear, that is usually caused by the temporal bone injury or develops after stapedoplasty. In some cases the condition is complicated by the oval window fistula [5]. Several cases of pneumolabirynth have been reported in literature: the majority of cases were diagnosed in patients who had undergone primary stapedoplasty. The other causes are the stapes footplate fracture or dislocation, penetrating trauma, temporal bone injury, barotrauma, and cochlear implantation [6].

In the majority of reported cases, pneumolabirynth occurred within weeks or months after stapedectomy. This complication usually manifests itself as a combination of complaints of vestibular (dizziness, bouts of nausea) and cochlear disorders (rapid sensorineural hearing loss, ear fullness and tinnitus).

High-resolution CT is essential for the diagnosis of the pneumolabyrinth. Air in the inner ear and possible stapedial prosthesis displacement indicate the presence of pneumolabirynth [7].

There is no clear algorithm for treatment of the condition due to the small number of cases reported in literature. In patients, who experience rapid hearing loss and vestibular symptoms in the short-term postoperative period (from a few

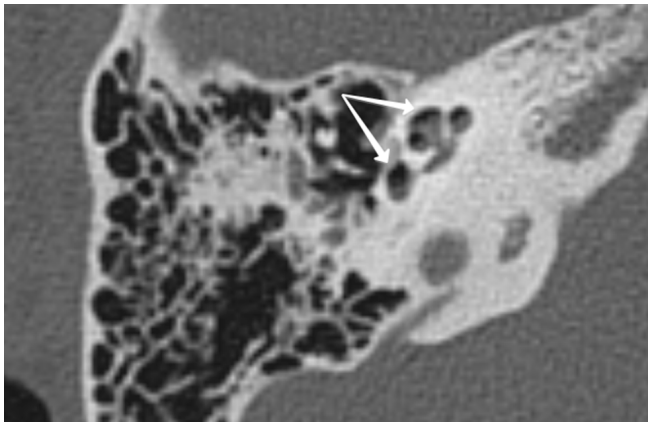

Fig. 6. CT scan performed during the early postperative period, right temporal bone of patient S. aged 37: the presence of air in the cochlea and vestibule

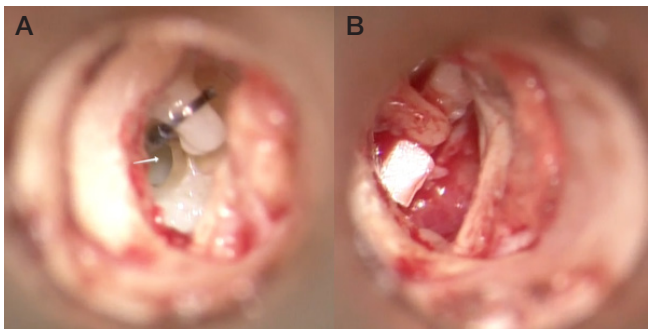

Fig. 7. A. The figure shows the stapedial prosthesis in the correct position that is fixed on the long process of the incus: the *arrow* marks the "open" fistula of the oval window, the "empty" vestibule. **B**. The figure shows the correct position of the autocartilage columella (sesamoid bone is located in the notch of the autocartilage distal end) at the final stage of the surgery

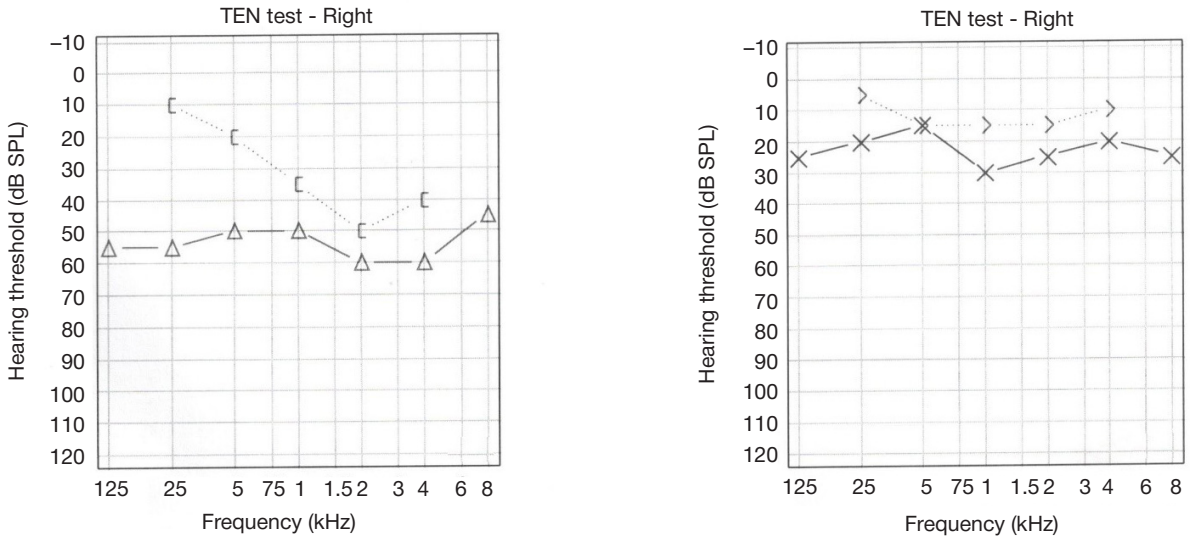

Fig. 8. PTA data of patient S., 37 years old, obtained in the short-term postoperative period

days to three months) after stapes surgery, the treatment strategy (conservative therapy vs. revision tympanotomy with mandatory elimination of the cause) is selected based on the diagnosed type and nature of injury [8]. In cases when pneumolabirynth occurs as a delayed complication of stapedoplasty (within months or even years after surgery), the decision on treatment strategy is less certain [9, 10]. In the reported cases of the stapes surgery postoperative complications, revision tympanotomy was mostly used that involved sealing the defects with autogenous tissue grafts: perichondrium of auricular autocartilage (as in cases presented in our article); ear lobe fat graft; temporal fascia graft [11]. The cases reported in the article confirm the efficiency of this method for surgical treatment of pneumolabirynth and repair of the defect (perilymphatic fistula) with the autogenous tissue graft (autogenous perichondrial graft). Vestibular symptoms disappeared after surgical treatment in almost all the reported cases, supposedly due to vestibular compensation.

Thus, the outcomes of the vestibular system adaptation after surgery are definitely positive, regardless of the lesion type and severity, or the type of previous intervention. In contrast to vestibular symptoms, the results related to hearing repair are not that clear. Thus, in the 1st reported case of delayed pneumolabirynth, we failed to restore hearing to the socially significant levels, despite the fact that improved hearing (by one degree) in the operated ear was observed. In the 2nd reported case of early pneumolabirynth, hearing results were more successful.

The recent review [11] proposed three prognostic factors to predict hearing improvement after correction of pneumolabirynth: bone conduction hearing test at the onset of the disease, time interval from injury to surgery, and traumatic damage to stapes.

In the reported cases, in was decided to perform surgery in patients K. and S. due to pneumolabirynth (confirmed by the temporal bone CT scans), rapid hearing loss, and profound vestibular disorder. Perilymphatic fistula, that enabled communication between the middle ear and inner ear allowing air to enter the vestibule, was found during revision surgery.

Air intrusion to the perilymphatic space due to perilymphatic fistula is one of the mechanisms of hearing loss. The transition of the complex hydrodynamic system of the inner ear from the "closed" to the "open" state, associated with partial substitution of perilymph with air, dramatically alters normal physiological and physical parameters of the labirynth causing the decrease in the cohlear microphonic response, complex action potential, and endocochlear potential [12]. Removal of air from the labirynth by replacing air with liquid together with closure of the defect bring the system of the inner ear to the situation of normal physiological communication. In some cases this results in improvement (or even restoration) of cochlear microphonic response amplitude and action potential, and, consequently, in improved hearing [6].

The cases reported in this article support the theory proposed by the authors who earlier addressed the issue of the development of pneumolabirynth and treatment strategy. Both patients had vestibular disorders and profound hearing loss that significantly improved after surgery in patient S. and improved, in patient K. (although did not get back to baseline levels). It should be remembered that in case of pneumolabirynth due to stapedial prosthesis subluxation the patient should be aware that the prosthesis removal with subsequent installation in the correct position may not improve hearing, but significantly improve or completely eliminate the vestibular symptoms [13, 14].

We believe that in the reported cases it is advisable to carefully remove the remaining footplate, remove the prosthesis to inject dexamethasone solution, and perform stapedoplasty with autocartilage on the vein. A total of 400 operations (stapedoplasty) are performed annually at the Department, and only two cases of pneumolabirynth have been identified within three years. In both cases, pneumolabirynth occurred due to patient non-compliance in the postoperative period, there was no prosthesis displacement (see the images obtained during surgery). According to our observations, the reported complications are not related to the type of prosthesis.

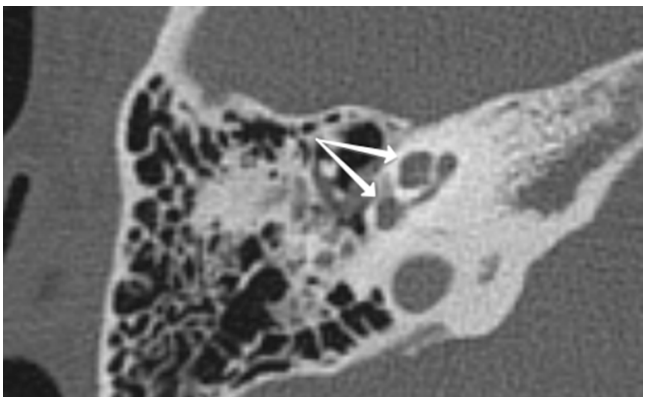

Fig. 9. CT scan performed during the short-term postperative period, left temporal bone of patient S. aged 37: the *arrows* indicate the cochlea and vestibule filled with liquid content

The cases reported in the article highlight the importance of surgical intervention for management of patients with pneumolabirynth and sensorineural hearing loss confirmed by audiograms.

CONCLUSION

Pneumolabirynth is a rare postoperative complication of stapes surgery. It should be suspected in patients with vestibulocochlear symptoms that emerge both in a short time and many years after stapedoplasty. In case of suspected pneumolabirynth it is necessary to conduct an examination in order to confirm the presence and circulation of air in the affected structures of the inner ear. The temporal bone CT is the best imaging tool used to confirm the diagnosis. Revision tympanotomy is recommended to patients diagnosed with pneumolabirynth, sensorineural hearing loss and/or constant dizziness.

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