

Case Report

Interdisciplinary Approach to Diagnosis and Treatment of Jugulotympanic Glomus Tumor with Dural Arteriovenous Fistula: Case study

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Abstract

Pulsating tinnitus is a special form of ear noise synchronized with the heart rate and is characterized by a high probability of organic cause. Glomous tumors of the middle ear and jugular foramen are rare neuroendocrine neoplasms with pronounced vascularization. Dural arteriovenous fistulas can be manifested mainly by otological symptoms, which often leads to diagnostic errors. The combination of these pathologies is extremely rare.

Together with audiologists and neurosurgeons, we present the third known case of severe pulsating tinnitus in a 79-year-old patient caused by jugulotympanic glomus tumors associated with a dural arteriovenous fistula.

After conducting audiological examination and a comprehensive interdisciplinary examination, a decision to perform simultaneous endovascular treatment, including embolization of the glomus tumor and dural arteriovenous fistula.

Keywords: pulsating tinnitus, glomous tumor, dural arteriovenous fistula, hearing loss, tympanometry, Tinnitus Handicap Inventory, endovascular treatment, interdisciplinary approach.

1. Introduction

Pulsating tinnitus is a distinct clinical manifestation synchronized with the heartbeat, which carries a remarkably high probability of an underlying organic, primarily vascular, etiology. Timely and precise differential diagnosis of this condition is of paramount importance, as it frequently masks severe vascular malformations or neoplastic processes [1,2].

Diagnostic pitfalls during the initial otolaryngological and audiological assessments often result in mismanagement and potentially life-threatening complications. Consequently, establishing a robust interdisciplinary paradigm involving audiologists and neurosurgeons remains highly relevant for optimizing patient outcomes. Thus,

pulsating tinnitus is one of the clinical "red flags" in otoneurology and requires the mandatory exclusion of vascular and tumor causes [3-5].

Incidence. Glomus tumors (paragangliomas) of the middle ear and jugular foramen are rare, hypervascular neuroendocrine neoplasms [1,2]. Similarly, dural arteriovenous fistulas (DAVFs) are uncommon vascular lesions that frequently present with predominantly otological symptoms, mimicking benign ear pathologies and leading to diagnostic delays. Notably, the synchronous coexistence of both a jugulotympanic glomus tumor and a DAVF in a single individual is an extremely rare phenomenon in global clinical practice [6].

This case report presents exceptional clinical and scientific novelty, as it details only the third documented case worldwide of severe pulsating tinnitus secondary to a combined pathology—a jugulotympanic glomus tumor associated with a DAVF—in a 79-year-old patient. The uniqueness of this presentation is further underscored by the successful application of an aggressive interdisciplinary strategy,

culminating in simultaneous endovascular intervention. These combined pathologies were described in (Darvie et al., 2018) and (Arnautović, 1998) [7,8]. Performing single-session embolization of both the glomus tumor and the dAVF provided an immediate and comprehensive resolution of the symptoms, offering valuable insights into the management of such complex vascular anomalies.

2. Presentation of a clinical case

A 79-year-old patient complained of a constant throbbing noise in her left ear, progressive hearing loss, and headaches. The symptoms have been bothering the patient since 2016. She repeatedly turned to otorhinolaryngologists, while the detected changes were interpreted as an inflammatory disease of the middle ear (otitis media), and the therapy was ineffective.

Methods. The treatment was carried out by a multidisciplinary team that included an otorhinolaryngologist, an audiologist, and neurosurgeons. Informed consent was obtained in accordance with the legislation of the Republic of Kazakhstan. The audiological examination included otoscopy, speech and tonal audiometry, tympanometry, acoustic reflexes, and the Tinnitus Handicap Inventory (THI) questionnaire for tinnitus assessment. Radiological diagnostics included: CT angiography of the brain and subtraction angiography, which revealed a hypervascular glomus tumor and a dural arteriovenous fistula. Endovascular treatment was performed under general endotracheal anesthesia, transarterial and transvenous access. The embolizing material is Onyx-18/20. The control examination on the fifth day included otoscopy, audiometry, tympanometry, and a THI re-evaluation.

Otoneurological and audiological examination

Otorhinolaryngological status. The examination was performed using the endovideosystem on the Chammed Medstar otorhinolaryngological combine.

The external nose is well-shaped, nasal breathing is unobstructed. The nasal conchae are moderately subatrophied, and the nasal passages are free. The nasal septum is free of deformities.

Pharyngoscopy: the pharynx is symmetrical, the mucous membrane is not hyperemic, the palatine tonsils are not enlarged.

Laryngoscopy: The larynx is accessible to examination, the mucous membrane is pink, the vocal folds are gray, and they close completely when phonated.

AD: the mastoid area is painless. The external auditory canal is wide and clear. The eardrum is intact, with clear landmarks, and the light reflex is preserved.

AS: the mastoid area is painless. The external auditory canal is wide and clear. The eardrum is bright purple, hyperemic, the landmarks are blurred, and the light reflex is not visualized (Figure 1). There is marked pulsation in the anterosuperior, posterosuperior and anteroinferior quadrants.



Figure 1 - The left eardrum

Table 1 - Speech audiometry (acumetry) and tuning fork tests

Spoken language:		Whispered speech:	
Right - 6.0 meters;		Right - 6.0 meters;	
Left - 0.5 meters.		Left – not perceived.	
Weber's test is the lateralization of sound to the right.		Federici–Bing test: positive on the right, negative on the left.	

Pure-tone Audiometry. The study was performed on a Sentiero diagnostic audiometer (Path Medical).

AD: High-frequency sensorineural hearing loss was detected. The average hearing threshold was 32.5

dB, which corresponds to mild hearing loss according to the international classification.

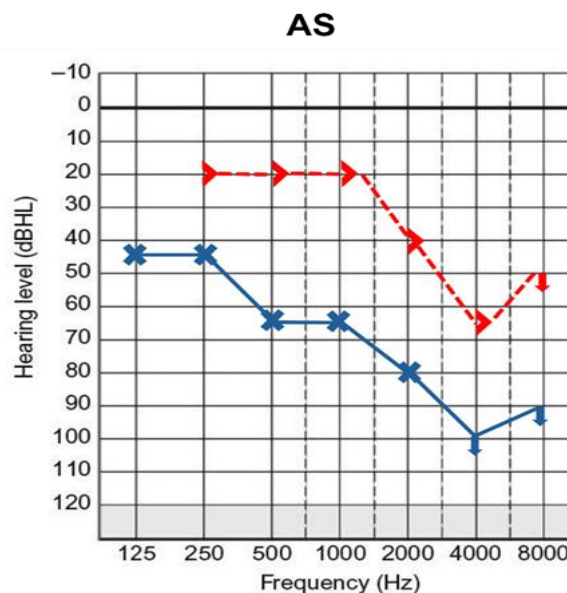
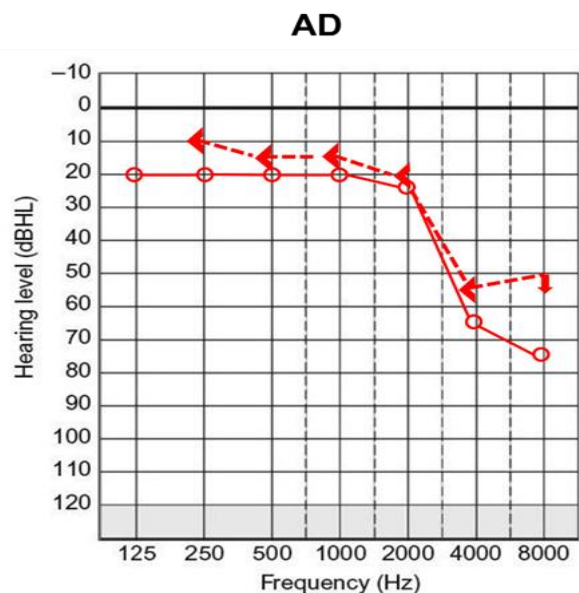


Figure 2 - Preoperative audiometry

AS: A significant air-bone gap of up to 50 dB was identified. The average hearing threshold was 70 dB, corresponding to severe hearing loss (Grade 4) according to the international classification (Table 1, Figure 2).

Impedance measurement

Tympanometry was performed on a Path Medical audiometer (Figure 3).

AD: type A tympanogram.

AS: type B tympanogram.

Acoustic reflexes: the Ipsi reflex is registered on the right, not registered on the left.

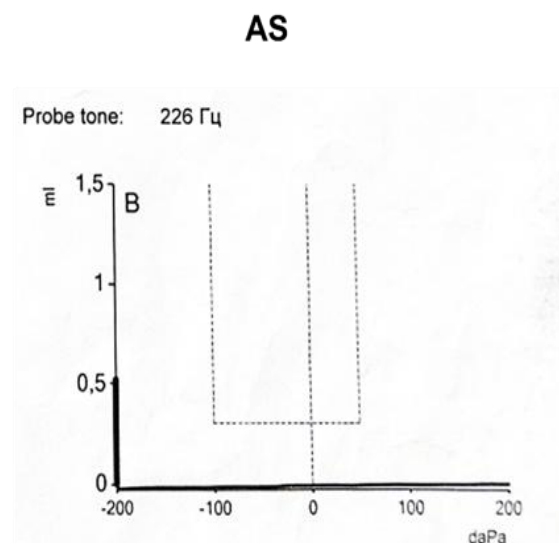
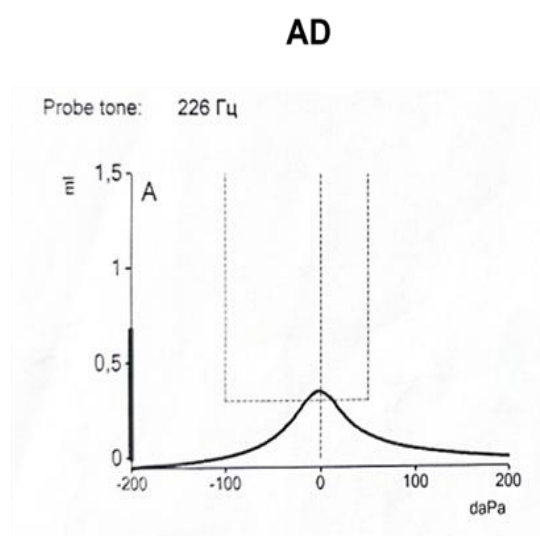


Figure 3 - Tympanometry

Justification of the choice and application of Tinnitus Handicap Inventory. The Tinnitus Handicap Inventory (THI) questionnaire was purposefully selected to assess the severity of tinnitus in the presented clinical case. The choice of this tool is due to the fact that tinnitus is a subjective symptom, the intensity of which does not always correlate with the results of an audiological examination and the degree of hearing impairment.

Unlike visual analog scales and short assessment tests, THI allows for a comprehensive assessment of the functional, emotional, and catastrophic aspects of tinnitus. This is of fundamental importance in patients with long-term pulsating tinnitus, in whom the severity of the symptom significantly affects daily activity, sleep, and mental and emotional state.

THI is a validated, reproducible and widely used tool in clinical practice and scientific research. The use

of a standardized questionnaire made it possible to objectify the patient's subjective complaints, identify a marked decrease in quality of life, and justify the need to review previously conducted diagnostic and therapeutic tactics.

In the presented case, a high score on the THI scale (60 points), corresponding to a severe degree of tinnitus, became a key argument in favor of conducting an extended vascular examination and interdisciplinary discussion of the clinical situation.

Radiology. CT angiography of the brain revealed hypervascular formation of the middle ear, jugular foramen, with red arrows (Figure 4).

An MRI scan was contraindicated in the patient due to the presence of metal structures in the body.

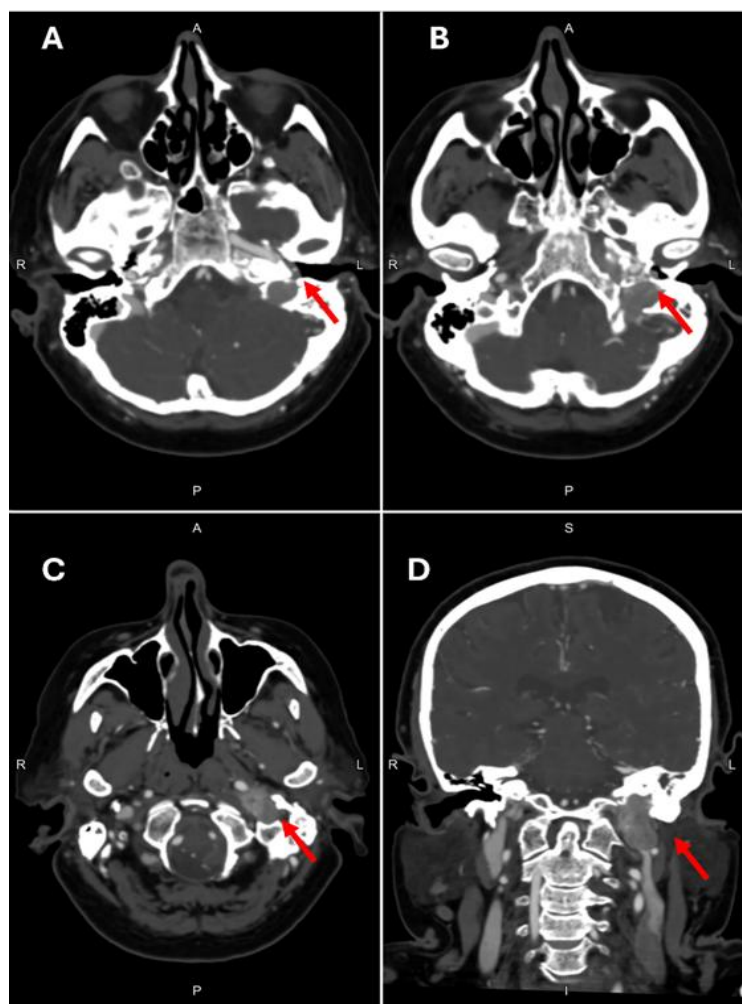


Figure – 4. CT scan of the brain with contrast

Treatment. The intervention was performed under general endotracheal anesthesia.

Vascular access - right common femoral artery, and a 6F introducer was installed.

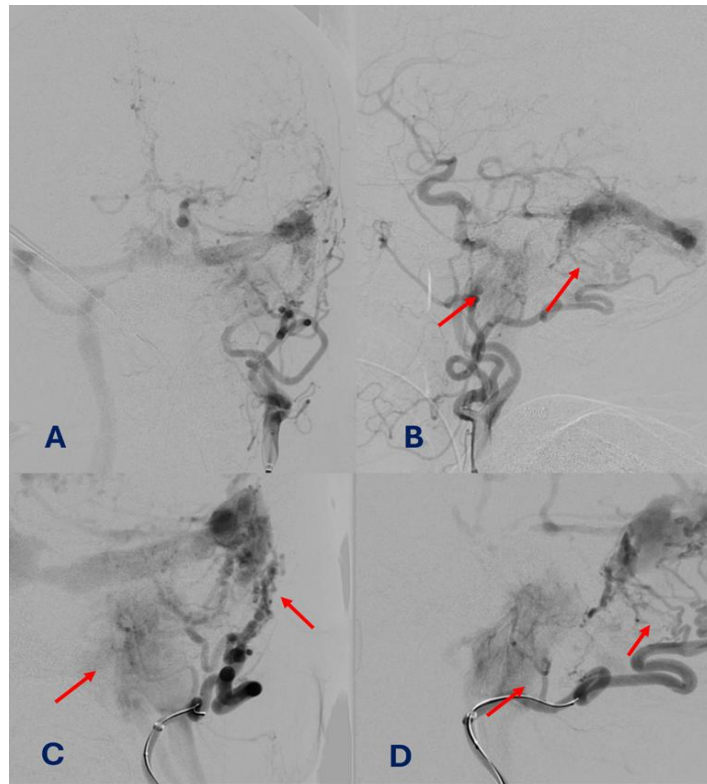


Figure 5 - Cerebral angiography in arterial and venous phases (A, B) visualized glomus tumor and DAVF (marked with red arrows). Selective angiography of the branches of the external carotid artery (C, D) revealed that feeder to the glomus tumor and DAVF arose from the branches of the hypertrophied occipital and meningeal arteries

According to the angiographic study, compression of the jugular bulb and the proximal internal jugular vein by a tumor was noted, which led to a violation of the physiological venous outflow (A). Venous drainage of the glomus tumor was carried out caudally into the distal parts of the internal jugular vein (marked with blue arrows on C, D). Venous outflow from the DAVF occurred in the transverse and sigmoid sinuses with

pronounced retrograde drainage, which corresponded to type IIb according to the Cognard classification.

Taking into account the high risk of venous hypertension and intracranial hemorrhagic complications, as well as a significant decrease in the patient's quality of life due to severe pulsating tinnitus, a decision was made on simultaneous endovascular treatment of both pathologies.

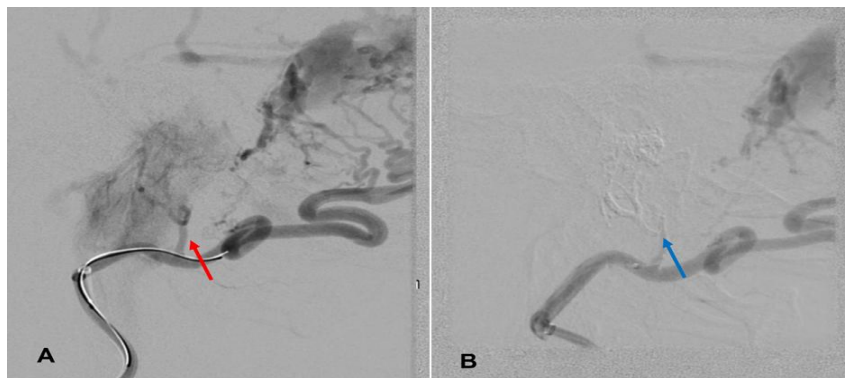


Figure 6 - The first stage. Endovascular embolization of a glomus tumor supplied with blood from a branch of the occipital artery (red arrow, A) was performed. The feeding vessel catheterized using a SONIC 1.5F/35 microcatheter. Onyx-18 liquid embolizate was introduced as an embolizing material. The control angiography after embolization is shown in the image (blue arrow, B)

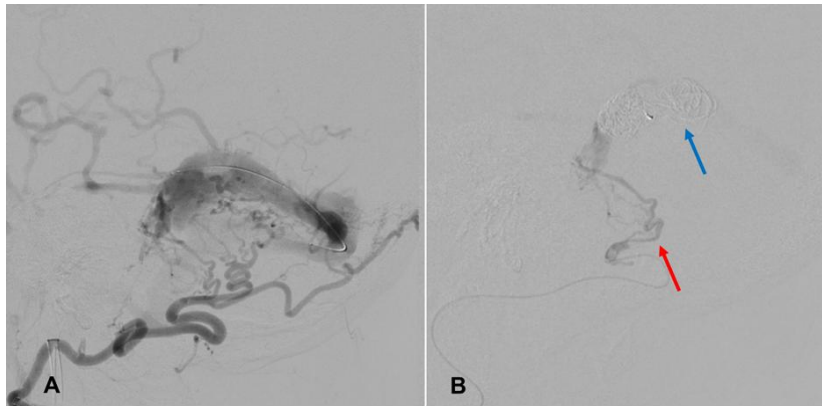


Figure 7 - The second stage. The right common femoral vein was punctured, and a guiding catheter was inserted into the internal jugular vein on the left. Transvenous shutdown of the transverse-sigmoid sinus was performed; the transition between the transverse and sigmoid sinuses was embolized by microspirals (blue arrow, B)

The third stage. Additionally, transarterial embolization of DAVF from the branches of the occipital artery was performed using a DMSO-

compatible Scepter 4.0 × 15 mm balloon catheter. Onyx-18 and Onyx-20 (red arrow, B) were used as embolizing materials.

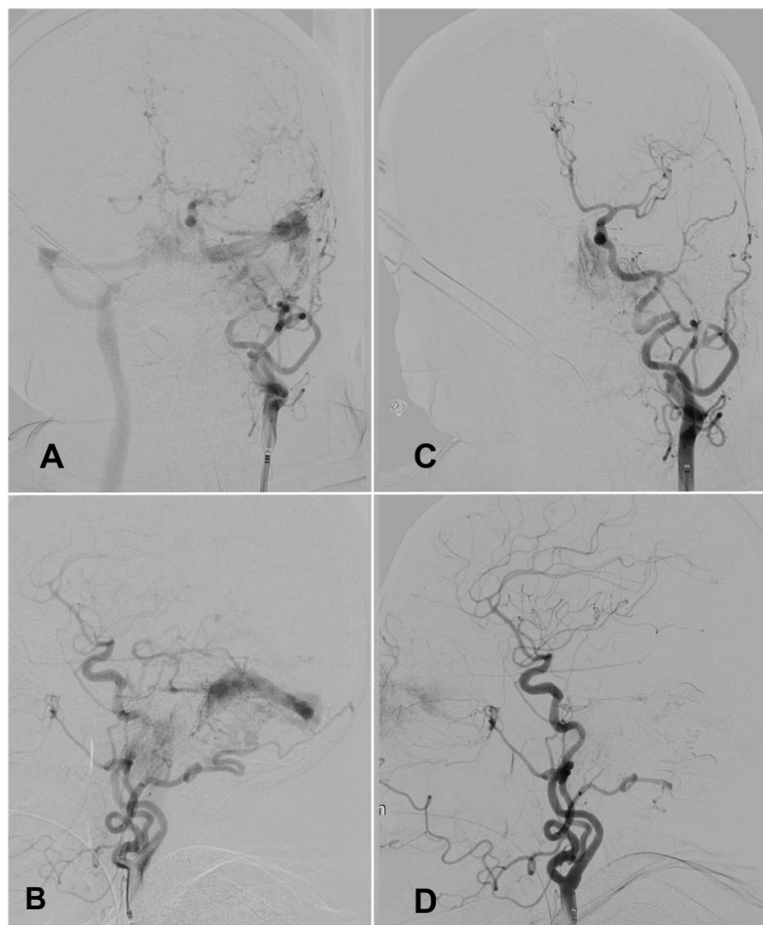


Figure 8 - Preoperative angiographic images (A, B). Postoperative angiographic images (C, D) confirm the complete shutdown of the pathological arteriovenous shunt and the absence of residual tumor blood supply.

After the intervention is completed, a pressure bandage is applied to the area of the postoperative access for a period of 24 hours

Results. 24 hours post-surgery, there was a partial disappearance of pulsating tinnitus, 120 hours after

surgery, and on the fifth day, the complete disappearance of pulsating tinnitus.

Post-treatment dynamics. On the fifth day after discharge from the hospital, the patient underwent a follow-up otoneurological and audiological examination.

Post-treatment otoscopy. AD: the mastoid area is painless. The external auditory canal is wide and clear. The eardrum is intact, with clear landmarks, the light reflex is preserved, and no pathological changes are detected.

AS: the mastoid area is painless. The external auditory canal is wide and clear. The eardrum is moderately hyperemic, the landmarks are distinguishable, and the light reflex is partially visualized. The pulsation of the eardrum is not detected.

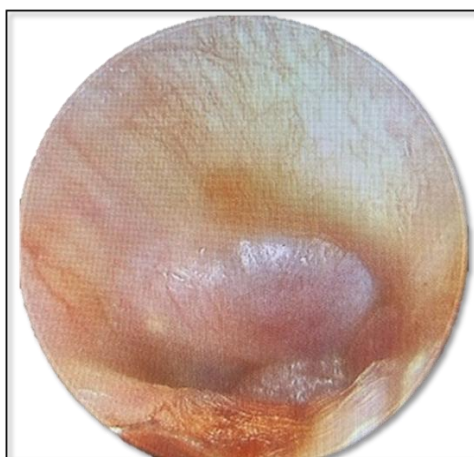


Figure 9 - Postoperative otoscopic image

Tinnitus assessment. Repeated assessment of tinnitus using the Tinnitus Handicap Inventory (THI) questionnaire showed a significant decrease in the severity of the symptom. The final score was 10 points, which corresponds to a slight (Grade 1) degree of tinnitus and indicates a pronounced clinical effect of the treatment.

Audiological examination. According to the follow-up pure-tone audiometry:

AD: The average hearing threshold was 27.5 dB, reflecting an improvement in auditory function compared with preoperative values.

AS: The average hearing threshold was 81.25 dB, which corresponds to a persistent severe hearing loss.

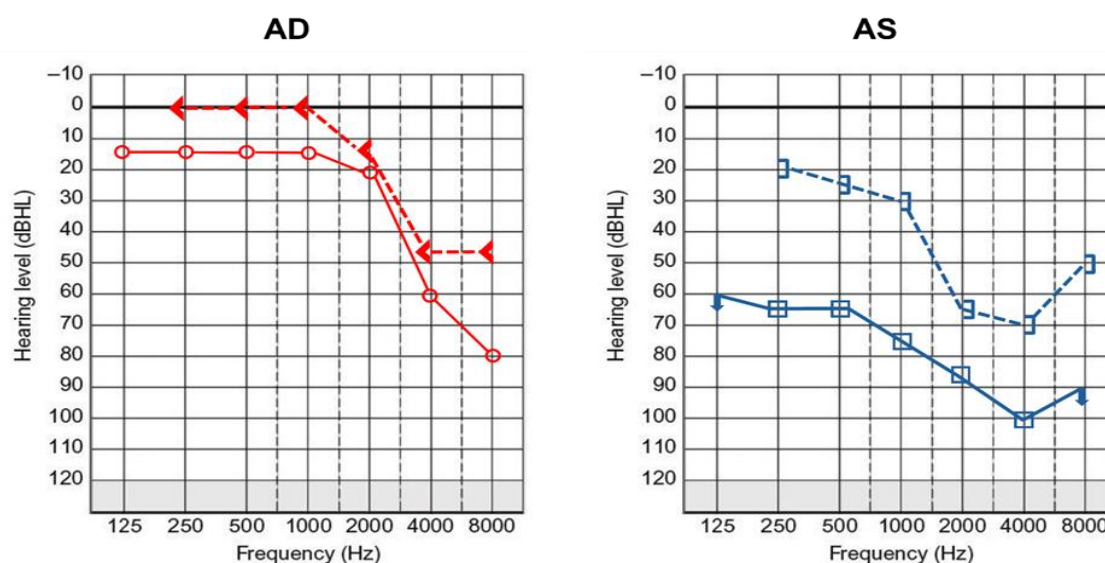


Figure – 10. Postoperative audiogram (day 5)

Tympanometry. The tympanometry data remained unchanged: AD: type A tympanogram. AS: type B tympanogram.

Quality of life. Despite the persistent hearing loss on the left, the patient noted a significant improvement in

the quality of life due to the complete resolution of pulsating tinnitus, normalization of sleep, and reduced in psycho-emotional stress.

4. Discussion

Given that glomous tumors of the jugular opening and tympanic cavity with dural arteriovenous fistulas are a rare combination, there are significant differences in treatment tactics and their effectiveness. In the case of Darvie et al. (2018), primary embolization of a glomous tumor did not lead to resolution of pulsating tinnitus, since this symptom was due not only to the volume of the tumor, but also to the associated dural arteriovenous fistula. Only subsequent occlusion of the dural arteriovenous fistula led to subjective relief [7].

In the case described by Arnautovic and co-authors (1998), the treatment of this pathology involved radical open surgery with preoperative embolization of the tumor and dural arteriovenous fistula. Such a combined approach to the treatment of highly vascular structures can result in total resection and radical surgery, but it is accompanied by the risk of cranial

nerve damage, postoperative neurological complications, and long-term rehabilitation [8].

Our experience shows that simultaneous endovascular embolization of both a glomous tumor and an associated dural arteriovenous fistula is a highly effective and safe tactic, especially in severe clinical syndrome of pulsating tinnitus. In contrast to step-by-step tactics, when tumor embolization does not lead to improvement, or radical surgery with high traumatism. Our technique has achieved complete regression of tinnitus already in the early postoperative period.

The results obtained emphasize the need for an individual interdisciplinary approach that combines audiological, otoneurological assessment, and treatment strategy planning, taking into account the angiographic characteristics of each component of pathology.

5. Conclusions

Pulsatile tinnitus is one of the clinical "alarm signals" in otoneurology and requires the mandatory exclusion of vascular and tumor causes. For several years, the patient's clinical symptoms were misinterpreted as inflammatory middle ear disease, leading to delayed diagnosis and a significant reduction in the patient's quality of life. An audiological assessment using the THI questionnaire and neuroimaging allowed us to objectively assess the severity of the symptoms and modify diagnostic and therapeutic approaches. Endovascular treatment of the patient resulted in complete regression of the pulsatile tinnitus in the early postoperative period. A comprehensive audiological examination and interdisciplinary approach allow for the timely identification of vascular causes of pulsatile tinnitus and significantly improve patients' quality of life.

A comprehensive audiological assessment and an interdisciplinary approach make it possible to identify

the vascular causes of pulsating tinnitus in a timely manner and significantly improve the quality of life of patients.

Ethical aspects. Informed voluntary consent of the patient has been obtained.

Funding. This research received no external funding.

Conflict of Interest. The authors declare no conflict of interest.

AI Declaration. AI-based tools were used exclusively for language editing and stylistic improvement of the manuscript. The authors take full responsibility for the accuracy and integrity of the manuscript.

Author Contributions. Conceptualization: Zh.T., Z.U.; Literature search: G.K.; Writing – original draft: G.K.; Writing – review & editing: G.K., Zh.T., Z.U.; Supervision: Z.U.

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Дуральді артериовенозды фистуламен ұштасқан югулотимпанды гломусты ісікті диагностикалау мен емдеудегі интердисциплинарлық тәсіл: Клиникалық жағдай

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Түйіндеме

Тамыр секілді соғатын тиннитус — бұл жүрек соғысының жиілігімен үндескен құлақ шуылының ерекше түрі, ол органикалық себептің болуының жоғары ықтималдылығымен сипатталады. Ортаңғы құлақ пен яремдік тесіктің гломусты ісіктері айқын васкуляризациясы бар сирек кездесетін нейроэндокриндік жаңа түзілімдерге жатады. Дуральді артериовенозды фистулалар негізінен отологиялық белгілермен көрінуі мүмкін. Бұл жиі диагностикалық қателіктерге әкеледі. Аталған патологиялардың бірге ұштасуы өте сирек кездеседі.

Аудиологтармен және нейрохирургтармен бірлесе отырып, біз дуральді артериовенозды фистуламен ұштасқан югулотимпанды гломусты ісіктен туындаған 79 жастағы емделушідегі ауыр, бүлкілдейтін тиннитустың әлемдік тәжірибедегі белгілі үшінші клиникалық жағдайын ұсынамыз.

Аудиологиялық зерттеу мен кешенді интердисциплинарлық консилиум жүргізілгеннен кейін гломусты ісік пен дуральді артериовенозды фистулань эмболизациялауды қамтитын бірізеттік эндоваскулярлық емдеу жүргізу туралы шешім қабылданды.

Түйін сөздер: тамыр секілді соғатын тиннитус, гломусты ісік, дуральді артериовенозды фистула, есту қабілетінің төмендеуі, тимпанометрия, тиннитус ауырлығының сауалнамасы, эндоваскулярлық емдеу, интердисциплинарлық тәсіл.

Междисциплинарный подход к диагностике и лечению югулотимпанальной гломусной опухоли в сочетании с дуральной артериовенозной фистулой: Клинический случай

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Резюме

Пульсирующий тиннитус представляет собой специфическую форму ушного шума, синхронизированную с частотой сердечных сокращений, которая характеризуется высокой вероятностью наличия органической причины. Гломусные опухоли среднего уха и яремного отверстия относятся к редким нейроэндокринным новообразованиям с выраженной васкуляризацией. Дуральные артериовенозные фистулы могут манифестировать преимущественно отологическими симптомами, что часто приводит к диагностическим ошибкам. Сочетание данных патологий встречается крайне редко.

Совместно с аудиологами и нейрохирургами мы представляем третий известный в мировой практике клинический случай тяжелого пульсирующего тиннитуса у 79-летнего пациента, вызванного югулотимпанальной гломусной опухолью в сочетании с дуральной артериовенозной фистулой.

После проведения аудиологического обследования и комплексного интердисциплинарного консилиума было принято решение о проведении симультанного (одномоментного) эндоваскулярного лечения, включающего эмболизацию гломусной опухоли и дуральной артериовенозной фистулы.

Ключевые слова: пульсирующий тиннитус, гломусная опухоль, дуральная артериовенозная фистула, тугоухость, тимпанометрия, опросник выраженности тиннитуса, эндоваскулярное лечение, междисциплинарный подход.