


A rare case of external ear canal stenosis caused by COVID-19-related arterial thrombosis

Ear, Nose & Throat Journal
2022, Vol. 0(0) 1–5
© The Author(s) 2022
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/01455613221097198
journals.sagepub.com/home/ear


Makoto Hosoya, MD, PhD¹, Ayumi Minami, MD¹, Takanori Nishiyama, MD, PhD¹, Nobuyoshi Tsuzuki, MD¹, Takeshi Wakabayashi, MD¹, and Naoki Oishi, MD, PhD¹

Abstract

Thrombosis is a characteristic symptom of coronavirus disease 2019 (COVID-19). Here, we present a case of external ear stenosis caused by arterial thrombosis after COVID-19 infection. To the best of our knowledge, this is the first report of external ear stenosis related to COVID-19. A 62-year-old man presented with left hearing loss. The patient had a history of hospitalization for COVID-19 treatment 11 months prior to visiting our hospital. He had been experiencing ear fullness and tinnitus after COVID-19 treatment. Physical examination revealed severe left external ear canal stenosis with a subcutaneous mass. Surgical removal of the subcutaneous mass was performed. Histopathological analysis revealed that a subcutaneous thrombosis caused the external ear canal stenosis. This case describes an unusual case of external ear canal stenosis after COVID-19. Clinical and pathological findings indicate that COVID-19 affected the external ear canal. In addition, histopathological results confirmed the formation of arterial thrombosis in the temporal bone region after COVID-19 treatment. This case shows the broad range of body sites that can be involved with thrombotic events with COVID including the subcutaneous tissue around the outer ear. This observation would be helpful in investigating or explaining the various otological symptoms of COVID-19.

Keywords

COVID-19, external auditory canal, external ear, otology

Introduction

Coronavirus disease 2019 (COVID-19) can cause various systemic complications. In the field of otorhinolaryngology, taste and olfactory disorders are well-known complications of COVID-19. While it is relatively rare to compare taste and olfactory disorders, several complications of COVID-19 that cause hearing loss have been reported.

It has also been confirmed that the SAR-CoV-2 virus can be detected in the middle ear cavity and mastoid cavity of patients with COVID-19¹. In addition, according to some reports, SAR-CoV-2 virus can cause viral otitis media, suggesting that acute otitis media can accompany COVID-19,² although the frequency is unknown. Other cases of acute otitis media have been reported as the initial symptom leading to the diagnosis of COVID-19.³ Regarding the effect of COVID-19 on the inner ear, there have been several case reports of acute

sensorineural hearing loss, including a case of viral meningitis that led to cochlear implantation.⁴ A case of sensorineural hearing loss with bilateral inner ear hemorrhage has also been reported.⁵ There has also been a report that acute sensorineural hearing loss was the first symptom of COVID-19.⁶

Despite the association of COVID-19 with middle and inner ear symptoms, there have only been a few reports about

¹Department of Otolaryngology-Head and Neck Surgery, Keio University School of Medicine, Tokyo, Japan

Received: April 4, 2022; accepted: April 11, 2022

Corresponding Author:

Naoki Oishi, Department of Otorhinolaryngology-Head and Neck Surgery, Keio University School of Medicine, 35 Shinanomachi, Shinjuku-ku, Tokyo 160-8582, Japan.
E-mail: oishin@keio.jp



the effects of COVID-19 on the external ear, with only one previous study reporting an increase in the incidence of otitis externa in COVID-19 patients.⁷ Here, we report an extremely rare case of external ear canal stenosis caused by COVID-19-related thrombosis.

Case report

A 62-year-old man presented with left tinnitus and left ear fullness at our department. He had been treated for severe COVID-19 with acute respiratory distress syndrome (ARDS) at another hospital for four weeks, approximately 11 months before visiting our hospital. When he was diagnosed and treated with COVID-19 in the previous hospital, the computed tomography (CT) images showed a crazy-paving pattern in the bilateral lung. The peripheral blood test at that time showed hyperferritinemia (3344 ng/ml) and elevated soluble interleukin 2 receptor (sIL2R) levels (1099 U/ml). During hospitalization for COVID-19 treatment in another hospital, he was treated with extracorporeal membrane oxygenation (ECMO) for approximately two weeks. After COVID-19 treatment, he had been suffering from ear fullness and tinnitus, and he visited our department. He had been also suffering from erectile dysfunction after COVID-19 treatment.

Physical examination revealed bilateral ear stenosis, which was more severe on the left side (Figure 1 A-C). He had been pointed to no stenosis in both external ear canals when he visited other clinics prior to the COVID-19 infection. At this point, the subcutaneous mass (asterisk in B and C) made the external ear canal notably narrower on the left side. He had right sensorineural hearing loss and left mixed hearing loss.

There were no invasive mass lesions in the external ear canal, as seen in the computed tomography (CT) images (Figure 1 E-H), while microcalcifications were observed in the left ear canal where the subcutaneous mass was located (Figure 1 H). Magnetic resonance imaging (MRI) results revealed slightly enhanced structures of the external ear canal (Figure 1 I and J).

At this point, the cause of the patient's bilateral external ear canal stenosis could not be determined. We performed a resection for the external ear stenosis of the left ear to resolve ear fullness and widen the external ear canal to improve hearing. Subsequently, more severe stenosis was observed under general anesthesia (Figure 2 A and B). An incision was made in the outer ear canal. A spherical mass was encountered when the incision was made in the narrowest region (Figure 2C); however, it was easily dissected from the surrounding subdermal tissues. After resection, we placed the perichondrium harvested from the tragus on the surgical field, where the bony wall of the external canal was exposed, and fixed it with fibrin glue. External ear canal stenosis has been successfully resolved after surgery.

Histopathological analysis revealed that the organized thrombosis formed in the artery was composed mainly of

removed tissue (Figure 2 C-F). These histological findings helped rule out other causes of external ear canal stenosis, including neoplastic lesions or other benign tumors. He had no medical history of another thrombosis prior to his COVID-19 infection. The combined clinical and histopathological findings suggested an external ear canal stenosis due to the mass caused by a COVID-19-related arterial thrombosis of the external ear canal. During the 6-month follow-up, the patient's left external ear canal was wider than before the surgery (Figure 2 G and H).

Discussion

Several systemic complications of COVID-19 have been reported. Thrombosis is a systemic complication of COVID-19 and thrombosis of the lung or deep veins has received much clinical attention because of its high fatality.⁸ However, COVID-19 can also cause thrombosis in other organs from the thin veins to the thick arteries through its proinflammatory, milieu-mediated, hypercoagulable state characterized by micro- and macro-vascular thrombotic angiopathy.⁹ Clinically, several symptoms or biomarkers, such as ARDS, hyperferritinemia, and elevated sIL-2 levels, are known risk factors for thrombosis or coagulopathy caused by COVID-19.^{10,11}

It has been reported that arterial thrombosis has been observed in 4.4% of severe COVID-19 patients.¹² The anatomical distribution of these arterial thromboses was broad and they can be observed in the arteries, such as the limb arteries, cerebral arteries, and the great vessels (aorta, common iliac, common carotid, brachiocephalic trunk, coronary arteries, and superior mesenteric artery).¹² Central retinal artery occlusion has also been reported in COVID-19 patients.¹³ Thrombosis of superficial veins has also been reported.^{14,15} These venous thromboses have sometimes been observed as superficial thrombophlebitis.¹⁶

In this report, we present a case of histologically confirmed external auditory canal arterial thrombosis after COVID-19. There was no other coagulopathy or history of thrombosis in the patient, but this thrombosis developed after COVID-19 treatment. Therefore, we concluded that this external canal thrombosis is COVID-19-related symptoms. The patient was at high risk for coagulopathy and thrombosis with ARDS, hyperferritinemia, and elevated serum sIL-2 levels, all of which support this conclusion. His other clinical symptoms after COVID-19 treatment, erectile dysfunction, also suggested the existence of the systemic thrombosis in the patient.

To date, this is the first reported case of COVID-19-related external ear canal stenosis, to the best of our knowledge. Here, arterial thrombosis can be observed in the external ear canal. Moreover, histopathological results confirmed the formation of arterial thrombosis in the external ear canal after COVID-19 treatment. This observation indicated that thrombosis could be formed in the temporal bone region, similar to other organs

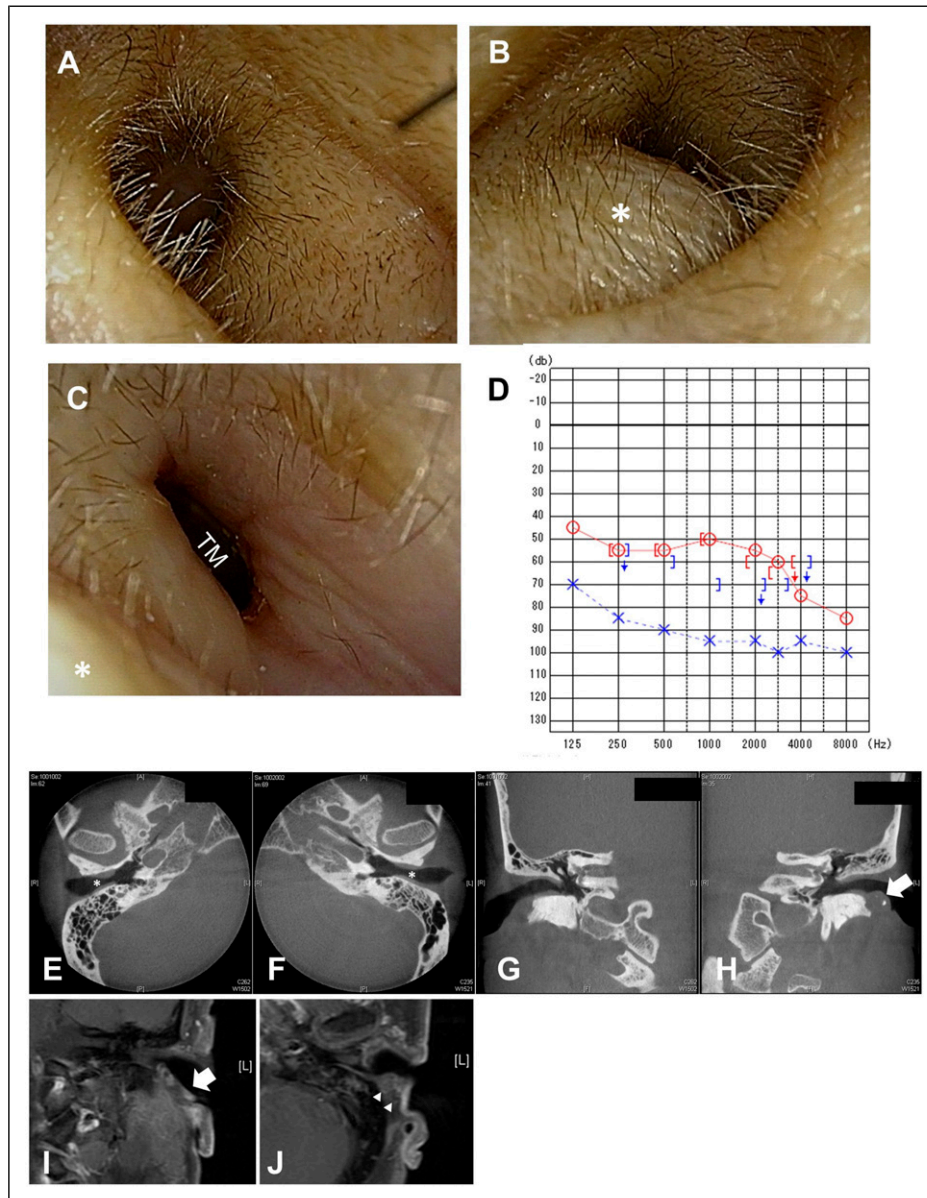


Figure 1. Physical examination findings of the external ear canal, hearing test on the first visit, and images from computed tomography (CT) scans and magnetic resonance imaging (MRI). (A–C) Bilateral external ear canal stenosis was observed (A: right, B and C: left). The subcutaneous mass (asterisk in B and C) made the external ear canal notably narrower particularly on the left side. (D) Audiometry shows the sensorineural hearing loss in the right ear and mixed hearing loss in the left ear. (E–H) CT scan images show bilateral external ear canal stenosis (asterisks in E and F) without accompanying bony destruction. Microcalcifications were observed in the subcutaneous mass on the left ear canal (arrow in H). No specific findings were observed in the middle ear. (I and J) MRI revealed slightly enhanced structures of the external ear canal (arrow in I and arrowhead in J). TM: tympanic membrane.

after COVID-19. Arterial thrombosis in the temporal bone region may cause other otological symptoms of ischemia. Our observation regarding the formation of arterial thrombosis in the temporal bone region would be helpful in investigating or explaining the various otological symptoms after COVID-19 infections in the future.

Here, we present an extremely rare case of external ear canal stenosis caused by thrombosis in the external ear canal

related to COVID-19. Several otological complications of COVID-19 in the inner or middle ear have already been reported. However, our case indicated that COVID-19 could also cause external ear canal complications in a specific situation. This case shows the broad range of body sites that can be involved with thrombotic events with COVID including the subcutaneous tissue around the outer ear. It also suggests the importance of a careful examination of the external ear canal

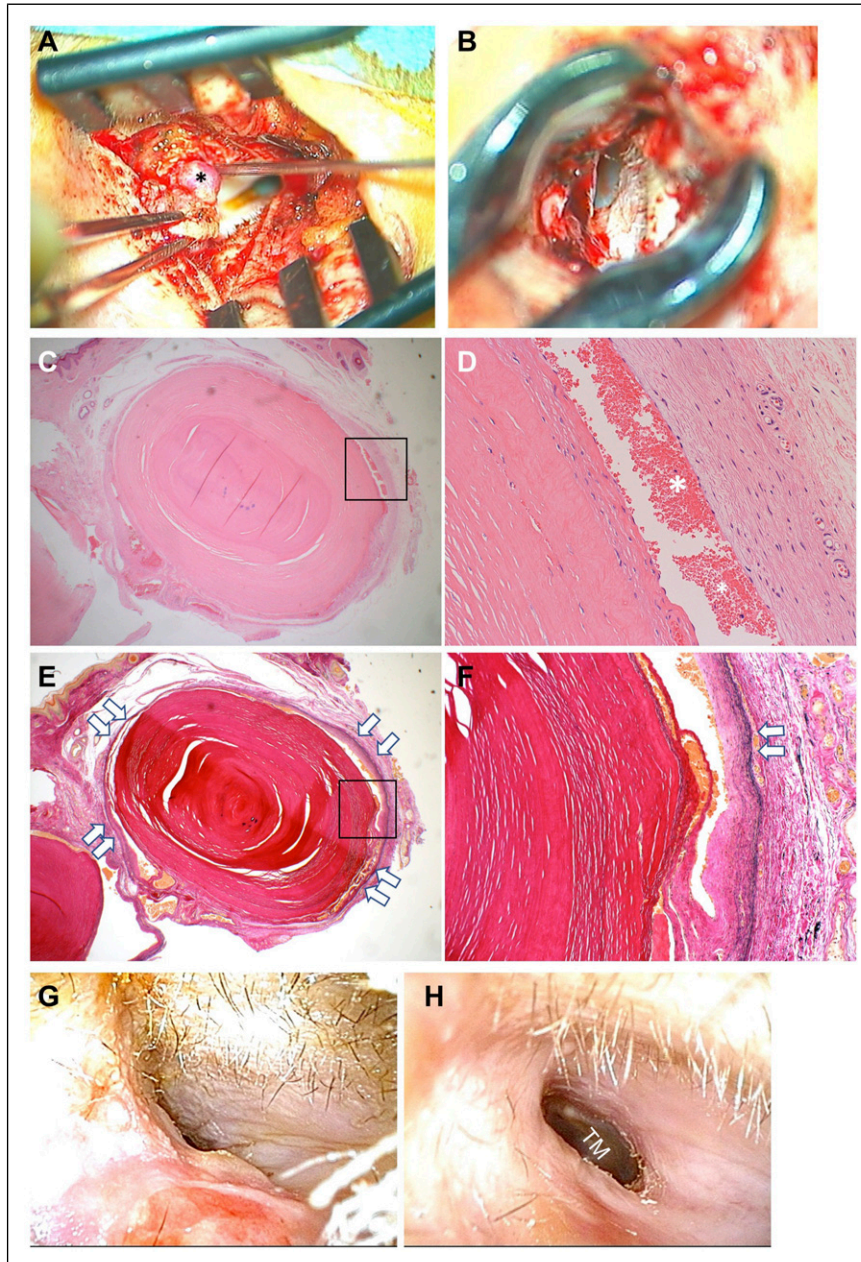


Figure 2. Intraoperative findings, histopathological findings, and local findings of the external ear canal of the left ear after surgery. (A and B) After the incision of the skin of the external ear canal, a spherical mass was observed where the tumor-like structure had been located (asterisk in A). The narrow external ear canal was relieved after resection of the mass. (C and D) Hematoxylin–eosin staining revealed the presence of red blood cells (asterisks in D) around an organized thrombus. (E and F) Elastica van Gieson staining showed the organized thrombus surrounding the elastic membrane (arrows in E and F). This observation indicated that this thrombus had been formed in the artery. (D) The high magnification image of the square shown in C. (F) The high magnification image of the square shown in D. (G) One-month post-operation. (H) Three-month post-operation. External ear canal stenosis has been successfully resolved after surgery. TM: tympanic membrane.

when patients consult regarding otological symptoms after COVID-19 since there might be narrowing or canal stenosis related to thrombosis caused by COVID-19.

Moreover, the histopathological findings in the case confirmed the formation of arterial thrombosis in the temporal bone region after COVID-19 treatment. This observation

would be helpful in investigating or explaining the various otological symptoms after COVID-19 infection.

Author Contributions

Makoto Hosoya, Ayumi Minami, Takanori Nishiyama, Nobuyoshi Tsuzuki, Takeshi Wakabayashi, and Naoki Oishi contributed to the

creation and revision of the manuscript. Makoto Hosoya, Ayumi Minami, Takanori Nishiyama, Nobuyoshi Tsuzuki, Takeshi Wakabayashi, and Naoki Oishi read and approved the final manuscript.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

Ethical Approval

Ethical approval to report this case was obtained from the ethics committee of the Keio University School of Medicine, approval number [20200033].

Statement of Human and Animal Rights

All procedures in this study were conducted in accordance with the ethics committee of the Keio University School of Medicine, approval number [20200033].

Statement of Informed Consent

Verbal informed consent was obtained from the patient for their anonymized information to be published in this article.

References

1. Frazier KM, Hooper JE, Mostafa HH, Stewart CM. SARS-CoV-2 Virus Isolated From the Mastoid and Middle Ear. *JAMA Otolaryngol-Head & Neck Surg.* 2020;146(10):964-966. doi:10.1001/jamaoto.2020.1922.
2. Fidan V. New type of corona virus induced acute otitis media in adult. *Am J Otolaryngol.* 2020;41(3):102487. doi:10.1016/j.amjoto.2020.102487.
3. Ye W, Xianyang L. A Novel Coronavirus Pneumonia Case Report From an Ear, Nose, and Throat Clinic. *The Laryngoscope.* 2020;130(5):1106-1107. doi:10.1002/lary.28655.
4. Degen C, Lenarz T, Willenborg K. Acute Profound Sensorineural Hearing Loss After COVID-19 Pneumonia. *Mayo Clin Proc.* 2020;95(8):1801-1803. doi:10.1016/j.mayocp.2020.05.034.
5. Chern A, Famuyide AO, Moonis G, Lalwani AK. Bilateral Sudden Sensorineural Hearing Loss and Intralabyrinthine Hemorrhage in a Patient With COVID-19. *Otology & Neurotology.* 2021;42(1):e10-e14. doi:10.1097/MAO.0000000000002860.
6. Kilic O, Kalcioğlu MT, Cag Y, Tuysuz O, Pektas E, Caskurlu H, Cetin F. Could sudden sensorineural hearing loss be the sole manifestation of COVID-19? An investigation into SARS-CoV-2 in the etiology of sudden sensorineural hearing loss. *Int J Infect Dis.* 2020;97:208-211. doi:10.1016/j.ijid.2020.06.023.
7. Mady OM, El-Ozairy HS, Wady EM. Increased incidence of otitis externa in covid-19 patients. *Am J Otolaryngol.* 2021; 42(3):102672. doi:10.1016/j.amjoto.2020.102672.
8. Di Minno A, Ambrosino P, Calcaterra I, Di Minno MND. COVID-19 and Venous Thromboembolism: A Meta-analysis of Literature Studies. *Seminars in Thrombosis and Hemostasis.* 2020;46(7):763-771. doi:10.1055/s-0040-1715456.
9. Loo J, Spittle DA, Newnham M. COVID-19, immunothrombosis and venous thromboembolism: biological mechanisms. *Thorax.* 2021;76(4):412-420. doi:10.1136/thoraxjnl-2020-216243.
10. Hanff TC, Mohareb AM, Giri J, Cohen JB, Chirinos JA. Thrombosis in COVID -19. *Am J Hematol.* 2020;95(12): 1578-1589. doi:10.1002/ajh.25982.
11. De Roubin V, Reynaud F, Coudroy R, Rodriguez M, Monseau G, Joly F, Bardin J, Boissier F, Chatellier D, Veinstein A, Robert R, Frat J-P, Thille AW. High risk of pulmonary embolism in acute respiratory distress syndrome related to COVID-19: an observational controlled-cohort study. *Annals of Translational Medicine.* 2021;9(8):630-630. doi:10.21037/atm-20-6796.
12. Cheruiyot I, Kipkorir V, Ngure B, Misiani M, Munguti J, Ogeng'o J. Arterial Thrombosis in Coronavirus Disease 2019 Patients: A Rapid Systematic Review. *Ann Vasc Sur.* 2021;70: 273-281. doi:10.1016/j.avsg.2020.08.087.
13. Ucar F, Cetinkaya S. Central retinal artery occlusion in a patient who contracted COVID-19 and review of similar cases. *BMJ Case Rep.* 2021;14(7):e244181. doi:10.1136/bcr-2021-244181.
14. Gianfranco Lessiani G, Andrea Bocatonda A, Damiano D'Ardes D, Giulio Cocco G, Giuseppe Di Marco G, Cosima Schiavone C. Mondor's Disease in SARS-CoV-2 Infection: A Case of Superficial Vein Thrombosis in the Era of COVID-19. *Eur J Case Rep Intern Med.* 2020;7(10):001803. doi:10.12890/2020_001803.
15. Hesam-Shariati N, Fatehi P, Fathi F, Abouzaripour M, Hesam Shariati MB. A case report of greater saphenous vein thrombosis in a patient with coronavirus (COVID-19) infection. *Tropical Diseases, Travel Medicine and Vaccines.* 2021;7(1):6. doi:10.1186/s40794-021-00131-9.
16. Demirbaş A, Elmas ÖF, Türsen Ü, Atasoy M, Lotti T. Superficial thrombophlebitis in a patient with COVID 19: Heparin treatment after evaluation of D-Dimer. *Dermatol Ther.* 2020;33(4): e13768. doi:10.1111/dth.13768.