

CASE REPORT

Clinical features of otogenic cerebral sinovenous thrombosis: Our experience and review of literature

Meherzi Abir | Gdissa Amal | Bellakhdher Mouna | Omri Malika |
Ben Sghaier Habiba | Houas Jihen | Kermani Wassim | Abdelkefi Mohamed

Department of Otorhinolaryngology,
Faculty of Medicine, University of
Sousse, Farhat Hached Hospital,
Sousse, Tunisia

Correspondence

Meherzi Abir, Department of
Otorhinolaryngology, Faculty of
Medicine, Farhat Hached Hospital, 33
Nesrinestreet, Jawhara city, Sousse,
Tunisia.

Email: abir-meherzi@hotmail.fr

Abstract

Otogenic cerebral sinovenous thrombosis is an intracranial complication secondary to otogenic disease; it is rare but could be a life-threatening condition. Its management is always challenging. This study aimed to focus on clinical features, on diagnosis of this pathology and to review the most controversial aspect of management of otogenic cerebral sinovenous thrombosis. We reviewed retrospectively 10 inpatients treated with cerebral sinus thrombosis secondary to otitis between 1995 and 2020. Ten inpatients (eight males and two females) with ages ranging from 11 to 77 years were diagnosed with otogenesis sinus thrombosis. The most commonly reported symptoms were headaches and otalgia. Five patients had mastoiditis, and 7 of the 10 patients had other concurrent complications: cerebellar abscess (three patients) and extradural empyema in two patients, retropharyngeal abscess in one patient, and meningitis in one patient. All patients were treated with broad-spectrum antibiotherapy, anticoagulation was used to treat nine patients, and surgery was required in nine patients. Nine patients had satisfactory resolution of their symptoms, but one patient had sequel as permanent loss of visual acuity. Because of the inconspicuous clinical presentation, and the probability of a fatal evolution, suspicion is essential for a proper diagnosis and suitable treatment.

KEYWORDS

anticoagulation, concurrent complication, management, MRI, otogenic sinus thrombosis, surgery

1 | BACKGROUND

Otogenic cerebral thrombophlebitis is a rare intracranial complication of otitis media in the modern age of antibiotics. It poses a danger that requires immediate diagnosis and urgent medical and surgical treatment. Complications are

now more likely to arise from chronic ear disease or cholesteatoma rather than acute otitis media.¹ The mortality has significantly decreased but still range from 5% to 10%.²

This study aimed to focus on clinical features, on diagnosis of this pathology and to review the most controversial aspect of management.

This is an open access article under the terms of the [Creative Commons Attribution-NonCommercial](https://creativecommons.org/licenses/by-nc/4.0/) License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.

© 2022 The Authors. *Clinical Case Reports* published by John Wiley & Sons Ltd.

2 | METHODS

We conducted a retrospective study on 10 patients with otogenic cerebral thrombophlebitis diagnosed and treated in patients admitted in the ENT Department of Farhat Hached Hospital over a period of the period of 25 years (1995 to 2020). Our series included 10 patients; they were eight men and two women with a sex ratio (M/F) = 4. The mean age was 35 years (11 years old–77 years old). Neurologic signs (headache) were present in five patients and fever was noted in three other patients. All patients were assessed by cerebral CT scan, three among them benefits from a cerebral MRI also. The diagnosis was confirmed by radiographic examination in all cases. 90% of patients were anticoagulated.

3 | RESULTS

A total of 10 inpatients were identified. The clinical of our patient was summarized in the descriptive [Table 1](#).

Four patients (40%) had poorly controlled diabetes. All patients had a history of otitis, either recurrent acute or chronic otitis media. The mean delay of consultation was 27 days (from 7 days to 3 months).

The most common symptoms were headache, reported in five patients (50%), otalgia in all patients, otorrhea in nine patients (90%), and fever in three patients (30%) ([Table 2](#)).

The etiology was related to an acute otitis media (AOM) in four cases (40%), cholesteatoma otitis media (COM) in three cases (30%), and necrotizing external otitis (NEO) in three cases (30%).

All patients had positive otoscopic findings, which included purulent ear discharge. Retraction pockets with cholesteatoma debris were present in two patients. Congested and retracted pars tensa was found in one patient ([Table 2](#)). The decreased visual acuity was found in one case (related to the extension of thrombophlebitis disease to the cavernous sinus).

All patients were assessed by cerebral CT scan with contrast (contrast-enhanced computed tomography CECT) of the brain and temporal bones. The “empty delta sign” (central non-enhancing clot surrounded by enhancing dural sinus wall) which is related to the presence of thrombus which has been objectified in seven patients (70%) ([Figure 1A,B](#)). Three patients (30%) underwent a Magnetic Resonance Imaging (MRI).

Thrombophlebitis disease was confined only to the lateral sinus in five patients (50%), extended to the internal

TABLE 1 Course of patients

Patient	Gender	Age	Diagnosis	Management
1	Male	11	AOM+thrombosis of the lateral sinus	Cefotaxim+ofloxacin
2	Female	34	COM+thrombosis of lateral sinus	Amoxicillin+clavulanic acid+Antro-mastoidectomy+anticoagulant therapy
3	Male	18	AOM+thrombosis of the lateral sinus and cavernous sinus+extradural empyema+meningitis+cerebral abscess	Cefotaxim+fosfomycine+metronidazole+Mastoidectomy+extradural empyema+anticoagulant therapy
4	Female	50	NEO+thrombosis of the lateral sinus	Ciprofloxacin+ceftazidime
5	Male	12	AOM+thrombosis of the lateral sinus+extradural empyema	Cefotaxim+ofloxacin+Mastoidectomy+extradural empyema evacuation+anticagulant therapy
6	Male	77	Fungal NEO (candida)+thrombosis of the lateral sinus+internal jugular vein	Voriconazole
7	Male	21	AOM+thrombosis of the lateral sinus+mastoiditis	Cefotaxime+ofloxacin
8	Male	38	COM+thrombosis of lateral sinus+internal jugular vein+mastoiditis+meningitis+cerebral abscess	Cefotaxime+vancomycin+metronidazole +Antro-mastoidectomy+incision of the sinus and evacuation of the clot+anticoagulant therapy
9	Male	59	Fungal NEO (mucormycosis)+thrombosis of the lateral sinus+retrpharyngeal abscess+cerebral abscess	Amphotericine B+anticoagulant therapy+the small size of the abscess does not require surgery
10	Male	30	COM+thrombosis of lateral sinus+internal jugular vein+mastoiditis+extradural empyema	Cefotaxime+fosfomycine+metronidazole+Antro-mastoidectomy+incision of sinus+evacuation of the clot+anticoagulant therapy

TABLE 2 Clinical findings

Signs	Number of patients
Fever	3
PDF (peripheral facial paralysis)	1
Exophthalmos with palpebral oedema	1
Torticollis	1
Cervical swelling	1
Retroauricular tenderness with swelling of the ear lobe	5
otorrhea	5
Retracted tympanic membrane	1
Narrowed external auditory canal	2

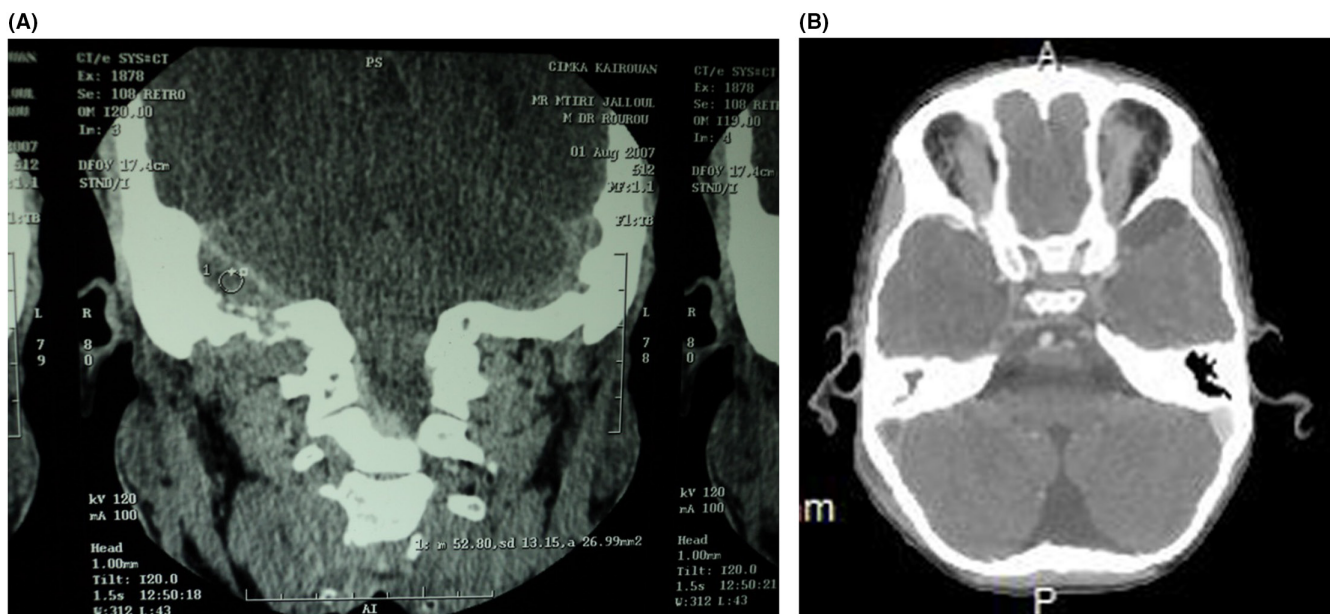


FIGURE 1 (A) Lateral sinus thrombosis. (B) Lateral sinus thrombosis

jugular vein in four patients (40%) (Figure 2), and extended to the cavernous sinus in one patient (10%) (Figure 3).

Regarding the patient with cavernous sinus extension, a CT scan revealed low-intensity cavernous sinus with bulging of its lateral edges, dilated ophthalmic veins, and bilateral exophthalmia, with the left eye having a more pronounced exophthalmia.

In three cases, the thrombophlebitis condition affected the right side, in six, the left side, and in one case, both sides. In four cases, the blockage was total and, in six cases, it was only partial.

Imaging had objectified other associated signs like mastoiditis which was identified in five patients (50%) (Figure 4), swelling neck in one case, and a retropharyngeal abscess in one case (Figure 5).

Other cranial complications were seen in 70% of patients, including cerebellar abscess (three patients), extradural empyema in two patients (20%), and meningitis in one patient (Figure 6).

Complete blood counts showed concentration of hemoglobin <10 g/dl in 2 (20%) patients, leukocytosis in 7 (70%) patients. All patients had normal plated counts. All patients had normal coagulation profiles.

Microbiologic cultures were produced from the middle ear of all patients and three of them had negative cultures. Among the positive cultures, *Pseudomonas aeruginosa* was isolated in two cases, *Proteus mirabilis* in one case, *Streptococcus* in one case, and *streptococcus pneumonia* in one case. Mycological cultures were positive in two cases: *lichenia corymbiform* in one case and *Candida Albicans* in the other case.

All patients received initially a broad-spectrum antibiotherapy, subsequently adapted according to the isolated germ. Antibiotic treatment lasted from 15 days to 3 months.

90% of patients were anti coagulated: six patients were treated with subcutaneous low molecular weight heparin (enoxaparin) for an average period of 60 days, and



FIGURE 2 Internal jugular vein thrombosis



FIGURE 4 Cerebral CT scan showing the mastoiditis's complication

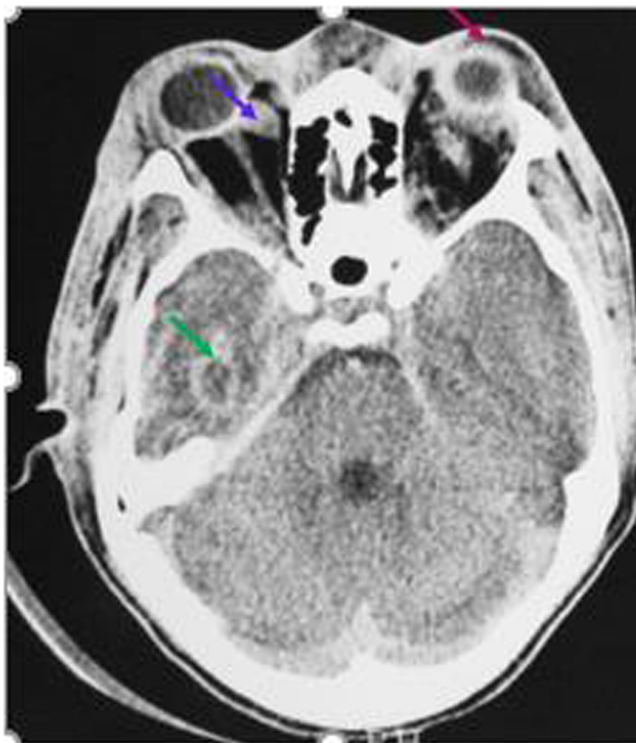


FIGURE 3 Cerebral CT scan illustrating thrombosis in cavernous sinus

three patients had intravenous unfractionated heparin for 15 days, then relayed with Sintrom (Acenocoumarol), for a mean period of 90 days.



FIGURE 5 Retropharyngeal abscess

Surgical management was performed in five cases (Table 1).

Except for one patient who experienced a loss of vision acuity, all patients made satisfactory recoveries, with all of their symptoms and problems completely resolved. In nine cases, the middle ear infection was under control. There was no mortality. One patient's thrombus significantly decreased, and four patients demonstrated recanalization. The follow-up period was 16 months long (range: 30 days to 36 months).

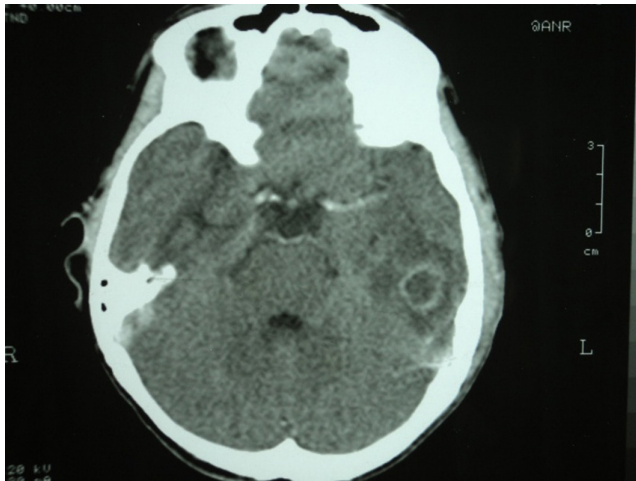


FIGURE 6 Cerebral empyeme

4 | DISCUSSION

The prevalence of cerebral sinovenous thrombosis has significantly decreased with the development of antibiotics, and in recent decades, there have been very few reports of it.³ With an incidence of 0.7 per 100,000 kids per year, this rare ailment has been more frequently recorded in the pediatric population³⁻⁵

Most studies^{2,4,6} have found a definite male predominance.⁷ In our series, we also discovered the similar prevalence.

Due to their anatomical proximity to the dural venous sinuses, the middle ear cavity and mastoid air cells are susceptible to thrombophlebitis brought on by infection and inflammation in these structures.⁸ The direct transmission of infection by erosive osteitis and retrograde thrombophlebitis are two pathogenic mechanisms of infection spread that have been proposed.^{9,10} The inflammatory process results in edema, a rise in local vascular pressure, and a hypercoagulable state. Consequently, it causes venous stasis and thrombosis to follow.¹¹

Nowadays, headaches were the most often reported symptom. The clinical presentation included otorrhea, hearing loss, and fever, according to Raja.K in her series "Otogenic Lateral Sinus Thrombosis: A Review of Fifteen Patients and Changing Trends in the Management".² This result was replicated in both Sherer's series and Bales' study.^{4,12} Otagia and vomiting were the commonest presenting features (63%) followed by fever (57%) and headache (43%) in Krishnan's study.¹¹

The primary signs and symptoms of cavernous sinus thrombosis included exophthalmos, decreased vision, ophthalmoplegia, ptosis, and palpebral edema along with headaches and fever.¹³⁻¹⁵ According to Zanoletti and coll, the existence of otogenic lateral sinus thromboses is not ruled out in the absence of the typical clinical indications

of mastoiditis.¹⁶ Fever and headache were both present in 50% of the individuals in our series.

The most widely cultivated microorganisms before the invention of antibiotics were B-hemolytic streptococcus and pneumococcus. Previously, *Pseudomonas* and *Proteus* species were widespread.¹

The profiles of microorganisms are evolving. A mixed flora, comprising *Pseudomonas*, *Proteus*, *Bacteroides*, *Staphylococcus*, *Enterobacteriaceae*, and other species, are frequently found in cultures of middle ear discharge. It could be often negative as a result of prior antibiotic use.² (1) Six patients (about 40%) in Raja's trial got negative cultures. *Proteus mirabilis* (four cases; 44%) and *Pseudomonas aeruginosa* (four cases; 44%) were the most common bacteria found in patients with positive cultures, followed by *Enterococcus faecalis* (one case; 11%) and *Escherichia coli* (one case; 11%). (2) Seven patients in our series had a positive culture. Five of them were associated with bacterial infections, two with mycologic infections, and one with an external ear infection. *Pseudomonas aeruginosa* was recovered in two of the positive bacterial cultures, *Proteus mirabilis* in one and *Streptococcus* in one.

The diagnosis of cerebral thrombophlebitis is based on radiographic imaging techniques. Contrast enhancing computed tomography of the head and neck is performed to investigate intra cranial complications of otitis media, especially the cerebral sinovenous thrombosis.^{5,9}

Lateral Sinus thrombosis may be diagnosed by the presence of the pathognomonic empty delta sign which consists of an empty triangle appearance created by the thrombus within the sinus surrounded by contrast-enhanced dura.^{5,9,14,17} In our study, the lateral sinus was the most common location in our patients. It was confined in 70% of cases and spread to the jugular vein in four cases and the cavernous sinus in only one case.

Cavernous sinus thrombosis may be initially explored with non-contrast CT of the head which can show subtle abnormalities such as bulging of the lateral margins of the cavernous sinus, heterogeneous filling defect, and engorgement of the superior and/or inferior ophthalmic veins. In addition to the above-mentioned signs, contrast-enhanced CT/MRI, shows the presence of asymmetric filling defects, thrombosis in the superior ophthalmic vein, other venous tributaries, dural venous sinuses, and cerebral veins.¹⁸ In our case, a CT scan was able to diagnose the thrombus in the cavernous sinus by showing low-intensity cavernous sinus, with bulging of its lateral margins, dilation of the ophthalmic veins, and bilateral exophthalmos more marked in the left eye.

CT scan can misdiagnose the thrombus pathology because of bone-related artifacts.¹ Therefore, M R I/M R V (magnetic resonance venography) is more sensitive in detecting this complication.²

To rule out other intracranial problems including nearby subdural empyema, cerebritis, or cerebral abscess, an MRI or MRV is helpful. Compared with CT scans, it can also reduce the danger of radiation exposure, especially for children.² The thrombus appears isointense on T1-weighted images and hypointense on T2-weighted images, with increased intraluminal sign intensity on both T1 and T2 sequences, and the MRI does not require contrast injection to show it.¹⁷ Because it is a non-invasive approach, the MRV continues to be the gold standard for identifying cerebral sinovenous thrombosis. It also has the advantage of precisely determining the patency of the central venous sinuses. Additionally, it allows for the differentiation between a slow venous flow and an occlusive thrombus and can be done in conjunction with cerebral MRI.^{11,17} In three cases in our study when the MRI identified the thrombus, the scanner was negative.

Additional intracranial complications must be investigated in the presence of cerebral sinus thrombosis. The high association of cerebral sinus thrombosis with other cranial complications is well documented.⁸

Concurrent complications were prevalent in 80% of cases in the pre-antibiotic period. Since the discovery of antibiotics, complications are now 20% less common. Meningitis, otitis hydrocephalus, internal jugular vein thrombosis, and intracranial abscesses were a few of the concurrent intracranial and extracranial problems.⁸

Syms and coll reported a group of patients who all had concomitant intracranial problems, including three patients with hydrocephalus and four patients with cerebral abscesses.¹⁹ Twelve of the thirteen patients in the Kaplan et al. study experienced concomitant problems, such as meningitis, cerebral abscesses, epidural abscesses, and development of the thrombus to the transverse sinus and the internal jugular vein.³

We have objectified 70% of the related cranial complications, including meningitis in one patient, extradural empyema in two patients, and cerebellar abscess in three patients.

Broad-spectrum intravenous antibiotics should be started at the earliest and must be adjusted later according to bacterial cultures. Antibiotics have led to a reduction of incidence of complications from 80% to 20%.¹ The duration of the antibiotic treatment ranged from four to eight weeks.² In our cases, we treated first with broad-spectrum antibiotics then adapted to the culture result.

Anticoagulation therapy's purpose in the management of LST is uncertain. In cases of cerebral sinovenous thrombosis, the clinician should consider the risks and advantages of anticoagulant medication. Although Au. JK et al.²⁰ showed a trend in the usage of anticoagulant treatment, they did not find a statistically significant difference.

Anticoagulation has evolved to have the benefit of stopping the thrombus from spreading to distal sinuses. In special instances of thrombus propagation, embolic events, and neurological alterations, it may therefore be indicated.^{1,21} The American Stroke Association advises Low molecular weight heparin (LMWH) for kids with Cerebral sinovenous thrombosis (CSVT) outside of the newborn period, even if there is proof of cerebral hemorrhage.^{5,22} It is recommended over other anticoagulants^{5,17} because it reduces the risk of long-term neurological sequelae, increases the rate of recanalization, and prevents thrombus propagation. However, it can also lead to thrombocytopenia, bleeding, hemorrhagic skin necrosis, and an increased risk of septic emboli.¹¹ Antibiotherapy combined with anticoagulation resulted in complete recovery in 74% of patients, with complete symptom relief and recanalization.²³

Nine patients recovered, and only one patient experienced complications after receiving anticoagulation, antibiotics, and surgical treatment for all of the patients.

Surgery is an essential part of the management of this entity, it improves prognosis. A better prognosis is ensured. However, controversies on the best surgical approach persist.¹¹ A cortical mastoidectomy is used successfully to treat noncholesteatoma ear disease. It confirms the diagnosis of L ST and allows the drainage of the initiating infection. A modified radical mastoidectomy is sufficient treatment for cholesteatomatous ears presenting acutely with cerebral sinus thrombosis.¹ Nowadays, routine ligation of the internal jugular vein is no longer performed. It is usually reserved for unresponsive cases with persistent septicemia, lung thromboembolism, and deep neck infection.^{2,9}

In our study, we realize a mastoidectomy isolated in four patients (40%), it was associated to an internal jugular vein ligation in one case, and to an extradural empyema evacuation in two patients (20%). We performed an antromastoidectomy with incision of the sinus and evacuation in two cases (20%).

5 | CONCLUSION

Otogenic cerebral sinovenous thrombosis is a rare complication of otitic pathology. It is associated with significant morbidity and mortality. In the era of antibiotics, classic clinical signs of mastoiditis (pain, swelling, and erythema posterior to the pinna) are not always present at the presentation. A clinical presentation might be subtle, which requires clinicians to maintain a high index of suspicion of this entity. Imaging (CT, MRI) is a key component in diagnosing and managing this complication. Treatment of otogenic CSVT consists of conservative surgery, antibiotics, and anticoagulation.

AUTHOR CONTRIBUTIONS

MA and GA charged of data collection, follow-up the case and writing the paper; BH and MO formulated the idea and supervised the manuscript preparation; BM and HJ participate in analyzing and interpreting data; KW and AM revising and approved the final version.

ACKNOWLEDGMENT

None.

FUNDING INFORMATION

None.

CONFLICT OF INTEREST

We have no competing interests.

DATA AVAILABILITY STATEMENT

All data and material of this case are available.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

REFERENCES

- Adhikari P, Guragain RPS. Lateral sinus thrombophlebitis: review of literature. *Intl Arch Otorhinolaryngol*. 2007;11:477-480.
- Raja K, Kumar Parida P, Alexander A, Surianarayanan G. Otogenic lateral sinus thrombosis: a review of fifteen patients and changing trends in the management. *Intl Arch Otorhinolaryngol*. 2018;22:208-213.
- Kaplan DM, Kraus M, Puterman M, Niv A, Leiberman A, Fliss DM. Otogenic lateral sinus thrombosis in children. *Intl J Pediatr Otorhinolaryngol*. 1999;49:177-183.
- Bales CB, Sobol S, Wetmore R, Elden LM. Lateral sinus thrombosis as a complication of otitis media: 10-year experience at the children's hospital. *Pediatrics*. 2015;123(2):709-713.
- Salloum S, Belzer K. Cerebral sinovenous thrombosis as a complication of otitis media. *Clin Case Reports*. 2019;7:186-188.
- Seven H, Ozbal AE, Turgut S. Management of otogenic lateral sinus thrombosis. *Am J Otolaryngol*. 2004;25(5):329-333.
- Fergoug I, Latroche M, Mehadji M. Thrombophlébite du sinus latéral d'origine otogène, prise en charge et pronostic. *Ann françaises d'Oto-rhino-laryngologie Pathol Cervico-faciale*. 2012;129:110.
- Viswanatha B, Naseeruddin K. Lateral sinus thrombosis in otology: a review. *Mediterr J Hematol Infect Dis*. 2010;3:27-33.
- Doyle KJO. Otogenic cavernous sinus thrombosis. *Otolaryngol Neck Surg*. 1991;104(6):873-877.
- Juilland N, Vinckenbosch P, et Richard C. Otite moyenne aiguë et complications à court terme. *Rev Médicale Suisse*. 2016;12:338-343.
- Krishnan M, Waljee H, Jesurasa A, De S, Sinha A, Sharma R. Clinical outcomes of intracranial complications secondary to acute mastoiditis: the Alder Hey experience. *Intl J Pediatr Otorhinolaryngol*. 2019;128:109675.
- Scherer A, Jea A. Pediatric otogenic sigmoid sinus thrombosis: case report and literature reappraisal. *Glob Pediatr Heal*. 2017;4:2333794X17738837.
- Bouslama M, Belcadhi M, Harzallah M, et al. Thrombophlébite du sinus caverneux d'origine otogène: a propos d'un cas. *J Tun ORL*. 2007;13:37-40.
- Gobron C, Guichard J, Chabriat H. Thrombose du sinus caverneux. *Sang Thromb Vaiss*. 2004;16:130-138.
- Babin E, Ndyaye M, Bequignon A, et al. Thrombose otogènes du sinus caverneux: à propos d'un cas. *Ann d'oto-laryngologie Chir cervico-faciale*. 2003;120:237-243.
- Zanoletti E, Cazzador D, Faccioli C, Sari M, Bovo R, Martini A. Intracranial venous sinus thrombosis as a complication of otitis media in children: critical review of diagnosis and management. *Intl J Pediatr Otorhinolaryngol*. 2015;79:2398-2403.
- Iseri M, Ayd N, Emre U, Alma A. Management of lateral sinus thrombosis in chronic otitis media. *Otol Neuro*. 2006;27:1098-1103.
- Bhatia H, Kaur R, Bedi R. MR imaging of cavernous sinus thrombosis. *Eur J Radiol Open [Internet]*. 2020;7:100226. doi:10.1016/j.ejro.2020.100226
- Syms MJ, Tsai PD, Holtel MR. Management of lateral sinus thrombosis. *Laryngoscope*. 1999;109:1616-1620.
- Au JK, Adam SI, Michaelides EM. Contemporary management of pediatric lateral sinus thrombosis: a twenty year review. *Am J Otolaryngol*. 2013;34(2):145-150.
- Ireo E, Gupta P, Dhanasekar G. Otogenic lateral sinus Thrombosis: a rare complication of chronic Otitis media. *Heighpubs Otolaryngol Rhinol*. 2017;1:46-52.
- Saposnik G, Barinagarrementeria F, Brown RD Jr, et al. Diagnosis and management of cerebral venous thrombosis: a statement for healthcare professionals from the American Heart Association/American Stroke Association. *Stroke*. 2011;42(4):1158-1192.
- Wong I, Kozak FK, Poskitt K, Ludernann JP, Harriman M. Pediatric lateral sinus thrombosis: retrospective case series and literature review. *J Otolaryngol*. 2005;34(2):79-85.

How to cite this article: Abir M, Amal G, Mouna B, et al. Clinical features of otogenic cerebral sinovenous thrombosis: Our experience and review of literature. *Clin Case Rep*. 2022;10:e06475. doi: [10.1002/ccr3.6475](https://doi.org/10.1002/ccr3.6475)