



Thrombosis of sigmoid sinus, transverse sinus, and internal jugular vein in chronic otitis media in 9-year-old girl: a case report

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Introduction and importance: Chronic otitis media can lead to dreadful intracranial complications, the most common being meningitis. A concomitant finding of thrombosis in more than one sinus with a cerebral vein is extremely rare. Septic sigmoid sinus thrombosis has an infectious origin and the treatment is debatable between antibiotics and surgery.

Case presentation: A case of 9-year-old female with prolonged symptoms of right sided ear discharge, fever, right sided neck pain, and vomiting. Examination revealed tachycardia and neck rigidity suggesting meningitis. The child developed shock, generalized seizure, and a persistent high grade fever in the course of hospital stay. Brain imaging revealed sigmoid, transverse, and internal jugular thrombosis. Blood culture grew both gram-positive and gram-negative bacteria, suggesting the infectious origin of the thrombus. The authors treated meningitis with cephalosporin and vancomycin at first, and then additional antibiotics to treat the septic sigmoid sinus thrombosis.

Clinical discussion: Sigmoid sinus thrombosis is a rare condition caused by infections, thrombophilia, head trauma, some types of cancer, and intravenous drug use. Cerebral vein or sinus thrombosis can lead to raised intracranial pressure and can cause fever, otalgia, headache, vomiting, cranial nerve palsies, papilledema altered mental status and may cause seizures, stupor, and coma. Prompt diagnosis by CT scan or MRI and prompt treatment with antibiotics are crucial.

Conclusion: Sigmoid sinus thrombosis with involvement of the transverse sinus and internal jugular vein is a rare complication of chronic otitis media, and should be suspected if a recurring fever with features of raised intracranial pressure is present in a child with chronic otitis media.

Keywords: case report, chronic otitis media, sigmoid sinus septic thrombus

Introduction

Chronic otitis media is a potentially serious disease because of its intracranial and extracranial complications^[1]. The most serious forms of intracranial complications are meningitis, otogenic brain abscess, and lateral sinus thrombosis, with a prevalence of 0.4–6.4%^[2,3]. Even though there has been a noticeable decrease in cases over a 15-year period, pediatric patients still experience intracranial complications from chronic otitis media (COM). This might be brought on by poor socioeconomic status, a lack of

HIGHLIGHTS

- Chronic otitis media can lead to dreadful intracranial complications, the most common being meningitis.
- A concomitant finding of thrombosis in more than one sinus with a cerebral vein is extremely rare.
- Sigmoid sinus thrombosis with involvement of the transverse sinus and internal jugular vein is a rare complication of chronic otitis media, and should be suspected if a recurring fever with features of raised intracranial pressure is present in a child with chronic otitis media.
- This is the first case report of its kind to be reported from Nepal.

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access to medical facilities, and widespread illiteracy in developing nations^[4,5]. Despite sigmoid sinus thrombosis being a rare complication, it can be fatal in both neonates and children. Sigmoid sinus septic thrombosis is the term used when an infectious cause is involved and it can be treated by prolonged use of broad spectrum antibiotics that cross the blood–brain barrier whereas surgical treatment by mastoidectomy is indicated as the treatment of choice by some literatures, especially if COM is the underlying cause. The use of oral anticoagulant agents or anticoagulants like heparin is even more debatable^[6–8]. Our article has been reported in line with Surgical CAse REport (SCARE) Criteria 2020^[9].

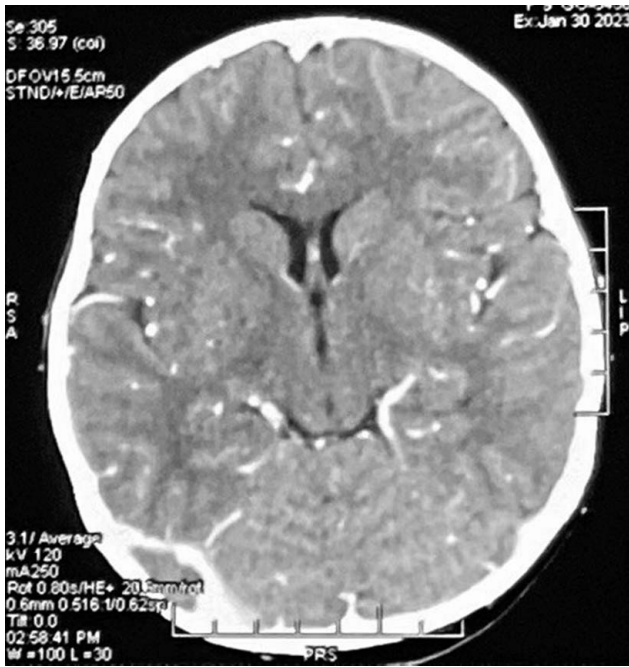


Figure 1. NCCT head showing transverse sinus thrombosis in a 9-year-old girl.

Case presentation

We present here a case of a 9-year-old female from Dharan, who is developmentally sound, fully immunized according to the National Immunization Program of Nepal, and who presented to the pediatric emergency with complaints of fever and right sided neck pain for 15 days along with headache and vomiting for 4 days. She also gives a history of right sided ear discharge for

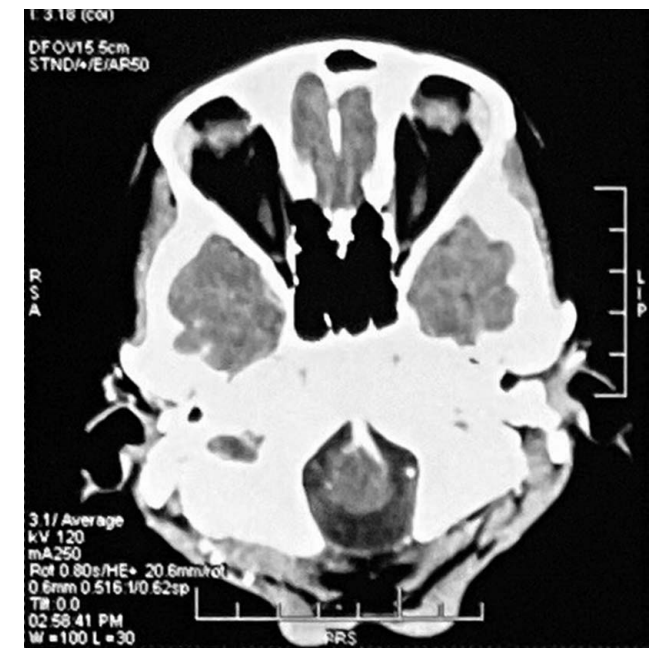


Figure 3. NCCT head showing sigmoid sinus thrombosis in a 9-year-old girl.

1.5–2 years, which was insidious in onset, scanty, foul smelling, and nonblood mixed.

Fever was acute in onset, continuous, maximum recorded up to 102°F (degree Fahrenheit), relieved after medication, associated with headache in the fronto-parietal region, acute in onset, moderate in intensity without chills or rigor. Fever was also associated with vomiting occurring after 10 days of the onset of fever, which was nonprojectile, nonbilious, and mixed with food

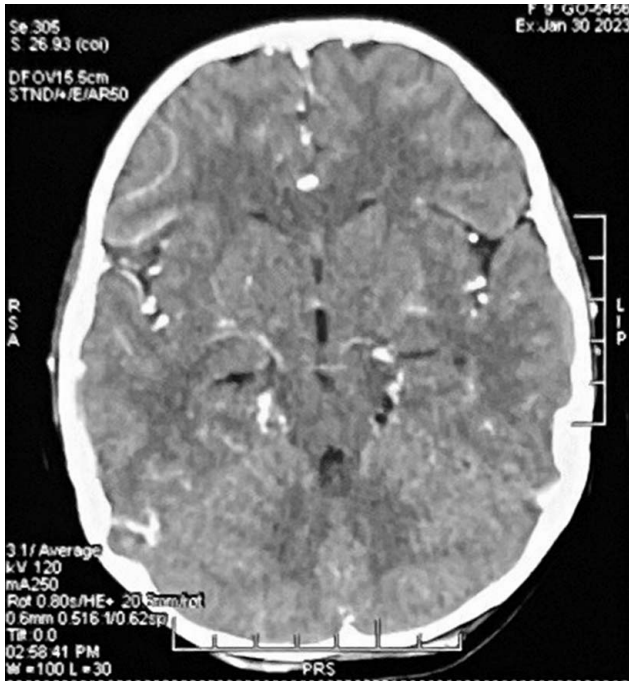


Figure 2. NCCT head showing sigmoid sinus thrombosis and transverse sinus thrombosis in a 9-year-old girl.

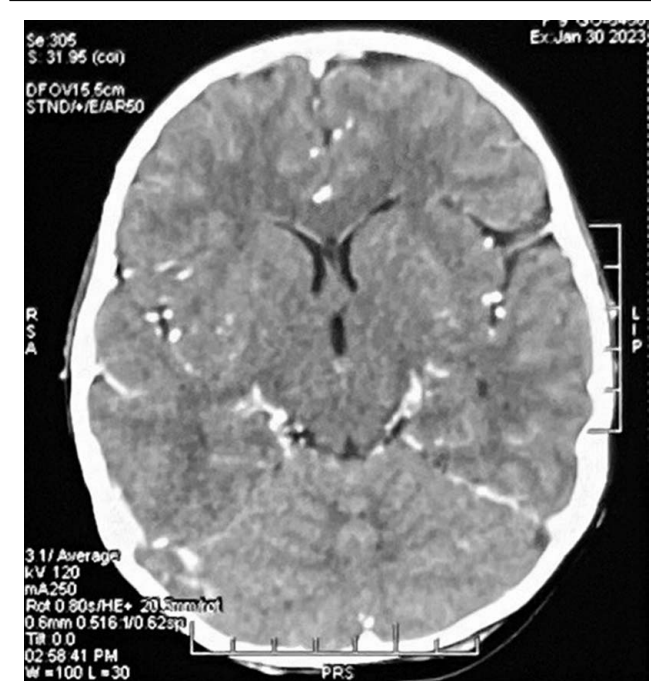


Figure 4. NCCT head showing right internal jugular vein thrombosis in a 9-year-old girl.

particles. She also complained of pain over right jaw and neck for 15 days. There is a history of tooth extraction done in the past due to dental caries. There is no history of tinnitus, ear fullness, vertigo, or ataxia. There is no history of loss of consciousness, any abnormal body movements, hematuria, hematemesis, or similar illnesses in the past. At the time of presentation, she was ill-looking with tachycardia (pulse rate: 140 bpm), normotensive, breathing normally (respiratory rate: 20 cycles/min), temperature: 101.8°F, and oxygen saturation of 98%. There was pallor, tender cervical lymphadenopathy on the right side and mild pedal edema. On neurological examination, neck rigidity was present. Other systemic examinations were unremarkable. A provisional diagnosis of meningitis secondary to right otitis media with cervical lymphadenitis with dental caries was made and empirical therapy was started with Ceftriaxone and Vancomycin. In view of the grade II papilloedema that was present in the child, a guarded lumbar puncture (under Mannitol infusion) was done. cerebro-spinal fluid (CSF) showed no cells, a normal glucose level (61 mg/dl), and normal protein (20 mg/dl). The CSF culture was sterile probably because of the use of prophylactic antibiotics. Two consecutive blood cultures were sent 16 h apart; the first grew *Proteus mirabilis* and the second grew *Enterococcus* species. However, the right ear swab culture was sterile.

ENT consultation was also done after, which the diagnosis was revised to meningitis with right chronic otitis media (squamous active) with right submandibular lymphadenitis with dental caries.

During the hospital stay, she responded well to antibiotics for the first few days, but on the second day, the patient developed shock. Noradrenaline was required upto 0.3 mcg/kg/min, which was gradually tapered off over 4 days. The patient also required a packed red cell transfusion on day 4 of admission. She also experienced one episode of generalized seizure, which were managed with phenytoin.

In view of persistent fever spikes, headache, and seizure, a contrast enhanced computed tomography head was done, which showed right-sided otomastoiditis with destruction of the lateral margin of the right sigmoid plate with thrombosis of the right transverse sinus as shown in Figures 1 and 2, thrombosis of the sigmoid sinus as shown in Figures 2 and 3, and thrombosis of a visualized part of the right internal jugular vein as shown in Figure 4. Based on blood culture sensitivity, antibiotics were upgraded to ceftazidime, vancomycin, and metronidazole. There was symptomatic improvement after a couple of weeks.

Clinical discussion

The presence of any one of the meningeal signs, such as stiffness or rigidity, meningeal irritation, the Brudzinski or Kernig sign, has a summary likelihood ratio of 4.50 and meningitis is less likely to occur when there are no meningeal signs^[10]. In our case, we made a clinical diagnosis of meningitis secondary to otitis media based on the presence of ear discharge, neck rigidity, fever, and vomiting. The most significant laboratory diagnostic test for meningitis is CSF analysis via lumbar puncture^[10]. The CSF findings were normal, which could be explained by the prior administration of antibiotics before presenting to the hospital.

The contrast enhanced computed tomography brain showed involvement of the right sigmoid plate with thrombosis of the

right transverse sinus, sigmoid sinus, and part of the right internal jugular vein, which was similar to a case reported in a 3-year-old boy with acute otitis media^[6]. However, our child had a history of chronic otitis media.

Sigmoid sinus thrombosis is a rare condition that is typically brought on by infections, thrombophilia, head trauma, some types of cancer, and intravenous drug use and cause in our case was infective. The most frequent causes of sigmoid sinus septic thrombosis (SSST), which has an infectious etiology, are acute or chronic otitis media^[7].

In a study conducted by Penido *et al.*^[8] in six patients of SSST, middle ear secretion culture was positive in three cases with the growth of *Enterococcus* spp., *Pseudomonas aeruginosa* and *P. mirabilis* and negative in the other three, which is explained by the use of broad spectrum antibiotics. In our case, the right ear swab culture was negative, which may be due to prior administration of antibiotics.

It is thought that a middle ear infection travels to the sigmoid sinus either directly or via the mastoid emissary vein, initially causing perivascular microabscesses and the continuation of the infectious process. The microabscesses eventually involve the venous system and result in an infected thrombus^[8].

Cerebral or venous thrombosis by preventing blood drainage from the brain tissue can lead to raised intracranial pressure and can cause fever, otalgia, headache, vomiting, papilledema, and seizure, which were evident in our patient. Depressed mental status, stupor, and coma are also the manifestation in these cases but were absent in our patient^[6]. Otorrhoea is also a frequent manifestation of SSST, which was present in our patient. However, postauricular edema, which is a frequent manifestation of SSST was absent in our case^[7].

For the patient's recovery in sigmoid sinus septic thrombosis, prompt diagnosis by CT scan or MRI and prompt treatment with broad spectrum intravenous antibiotics are crucial^[7]. While some advocates long-term use of broad spectrum antibiotics that cross the blood-brain barrier, some use surgical treatment by mastoidectomy (especially if chronic otitis media is the cause) for the treatment of SSST. The use of anticoagulants like heparin for the management of SSST is also an option, but it is very debatable^[8]. In our case, we tried 3 weeks of antibiotics for the treatment of SSST and no anticoagulants were given.

To our knowledge, concomitant thrombosis of the sigmoid sinus, transverse sinus and part of the internal jugular vein associated with COM and meningitis is rare. Pediatricians should have high an index of suspicion for sinus thrombosis in a child of COM presenting with an unremitting headache, fever with seizure.

Conclusion

Persistent otorrhoea for more than 2 weeks needs to be taken seriously in children. In a child of COM with prolonged symptoms of ear discharge, neck pain, and fever, brain imaging is crucial to rule out sinus thrombosis. It can decide the duration of therapy and the need for follow-up. A multidisciplinary approach involving an otorhinolaryngologist, pediatrician, and hematologist is required for managing such case.

Ethical approval

The study is exempt from ethical approval in our institution.

Literature review

| S.N | List of articles | References | Number of patients | Intervention | Results |
|-----|--|-------------------------------------|--------------------|---|--|
| 1. | Intracranial complications of CSOM in pediatric patients: a persisting problem in developing countries | Jain <i>et al.</i> ^[5] | 142 | Retrospective analysis. All patients underwent detailed ENT and neurological examination, PTA, HRCT temporal bone, Neurology/Neurosurgeon opinion sought, and mastoid exploration was done. MRI of brain, venography and neurosurgical exploration was done in required cases | The average age group was 13.8 years. The most common presentation was ear discharge followed by hearing loss, headache, fever, vestibular symptoms, neck stiffness and seizures. Atticoantral disease was seen in all the patients. Other signs were papilloedema, meningeal signs and cerebellar signs. The most frequent intracranial complication seen was brain abscess followed by meningitis, lateral sinus thrombosis and extradural abscess. Despite significant decline in the number of cases over years, CSOM continue to persist in pediatric patients which could be due to lack of access to health care, low socioeconomic status and illiteracy |
| 2. | Sigmoid sinus thrombosis as complication of otitis media in a 3-year-old boy: case report and review of the literature | Kalyva <i>et al.</i> ^[6] | 1 | The child underwent detailed history and examination. Initial blood work-up, fundoscopy, CT Head without contrast, CSF analysis, bilateral myringotomy with insertion of tympanostomy tube bilaterally, and subsequently underwent simultaneous bilateral mastoidectomy. VP shunt was also created for raised ICP | A 3-year-old boy with a history of recurrent acute otitis media presented with fever, otalgia, and headache for 3 days. He was initially treated partially with antibiotics for 1 month. Despite no signs of CNS involvement, clinical suspicion for CNS pathology guided the diagnostic work-up and led to the diagnosis of sigmoid sinus thrombosis. After that, the patient received intravenous antibiotics, anticoagulation therapy and underwent myringotomy, bilateral tympanostomy tube insertion, and mastoidectomy |
| 3. | Sigmoid sinus thrombosis associated to chronic otitis media | Penido <i>et al.</i> ^[8] | 6 | Retrospective analysis. MR angiography or arteriography | Study population was young patients, with mean age of 26 years, and extreme ages of 10 and 50 years. All of them had acute mastoiditis at the time of presentation, and four of them also had intracranial abscesses, three had meningitis, one had facial nerve paralysis, and another had an abducent nerve paralysis. Ear discharge, headaches, stiff neck, high fever, and malaise were the most common presentations. During image analysis, the lateral sinus thrombosis was found in all six patients. A mastoidectomy was always followed by a 3-month course of broad-spectrum antibiotics. Anticoagulation therapy was started in three cases, while anticoagulation was not necessary in the other three cases. Each case displayed a good clinical course without any after effects |
| 4. | Intracranial complications of otitis media: 15 years of experience in 33 patients | Penido <i>et al.</i> ^[4] | 33 | Retrospective study. Details of patients, otological diagnoses, audiometric tests, microbiologic data, neurological diagnoses, therapeutic approaches including medical and surgical options | The ages of the patients ranged from 6 months to 79 years, with a mean of 26.9 years. The otological diagnoses revealed chronic otitis media in 27 patients and six cases of AOM. Persistent fever, headache, and purulent otorrhea were the most common clinical symptoms of otitis media with intracranial complications. The common intracranial complication was otogenic brain abscess followed by meningitis, lateral sinus thrombosis, subdural empyema, meningococcal and epidural empyema. 58% patients had more than one complications. 27% patients had intratemporal complications associated with intracranial complication. Surgical intervention, that is Mastoidectomy in 26 patients and nonotological intervention was done in 7 patients. Neurological surgery was also done in 21 patients. At the 6-month follow-up, 66% of patients presented without sequelae, 24% presented with sequelae, and 9% died |

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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None.

Author contribution

All the authors were involved in manuscript preparation, review of literature, and final approval of manuscript.

Conflicts of interest disclosure

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Research registration unique identifying number (UIN)

Not done (no any new surgical technique or new equipment/technology used).

Guarantor

Ujjwal Kumar Shah.

Data availability statement

Yes the data analyzed during current study are publicly available, available upon reasonable request, or if data sharing is not applicable to this article.

Provenances and peer review

Not commissioned, externally peer reviewed.

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