


# Pediatric Otogenic Sigmoid Sinus Thrombosis: Case Report and Literature Reappraisal

Global Pediatric Health  
Volume 4: 1–8  
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DOI: 10.1177/2333794X17738837  
journals.sagepub.com/home/gph  


Andrea Scherer, MD<sup>1,2,3</sup>, and Andrew Jea, MD<sup>1,2,3</sup>

## Abstract

The purpose of this study was to review our experience with a single case of mastoiditis associated with sigmoid sinus thrombosis and increased intracranial pressure, and to review the experience of others through prior publications. We reviewed a case of a 6-year-old boy with an acute otitis media and mastoiditis, with associated ipsilateral sigmoid sinus and contralateral distal transverse sinus thrombosis. Based on the literature and our own experience, we conclude that most children with dural sinus thrombosis from acute otitis media and mastoiditis, in the setting of increased intracranial pressure, attain a good neurological outcome with a conservative neurosurgical approach. The mainstay of treatment seems to be appropriate antibiotic coverage and anticoagulation. Surgeries such as external ventricular drain, serial lumbar punctures, intracranial pressure monitor, and endovascular thrombectomy are reserved for patients with neurological deterioration despite maximal medical treatment.

## Keywords

otogenic, otitis media, mastoiditis, dural sinus thrombosis, otitic hydrocephalus, lateral sinus thrombosis

Received September 25, 2017. Accepted for publication October 3, 2017

## Introduction

Otogenic dural sinus thrombosis is a rare complication of acute otitis media and mastoiditis, especially in the era of exquisitely effective antimicrobial medications. Nonetheless, there are possible serious consequences with dural sinus thrombosis, including raised intracranial pressure (ICP) and hydrocephalus. Septic thrombosis of a dural venous sinus can result from direct extension of mastoiditis or septic granulation tissue overlying the sinus wall or from extension of infection through emissary veins that connect the mastoid with the sinus. The most frequently affected sinus is the sigmoid sinus because of its anatomic proximity and course to the mastoid part of the temporal bone. The thrombus may then propagate or embolize to the internal jugular vein or other dural venous sinuses.

Historically, sigmoid sinus thrombosis from mastoiditis included surgical drainage and removal of the infected thrombus.<sup>1,2</sup> However, recent studies showed that conservative surgical intervention with cortical mastoidectomy and eradication of the perisinus infection is effective, without drainage of the sinus and more

aggressive ICP management.<sup>2,3</sup> The indications for anticoagulation therapy are well defined in adults, but controversial in children.<sup>3,4</sup> Some authors cite the risks of anticoagulation in children, while others claim the risk of recurrent or propagated thrombosis if anticoagulation therapy is withheld.<sup>5-8</sup>

The purpose of this study was to review a difficult case of acute otitis media, mastoiditis, and sigmoid sinus thrombosis in a 6-year-old child. We treated this patient conservatively, and at the completion of treatment, a satisfactory outcome was obtained. We then compared and contrasted our management of this patient to 104 previous cases published in the literature.

<sup>1</sup>Riley Hospital for Children, Indianapolis, IN, USA

<sup>2</sup>Indiana University, Indianapolis, IN, USA

<sup>3</sup>Goodman Campbell Brain and Spine, Indianapolis, IN, USA

## Corresponding Author:

Andrew Jea, Section of Pediatric Neurosurgery, Riley Hospital for Children, 705 Riley Hospital Drive, Suite 1134, Indianapolis, IN 46202, USA.

Email: [ajea@goodmancampbell.com](mailto:ajea@goodmancampbell.com)



## Clinical Material and Methods

We retrospectively reviewed a case of acute otitis media, mastoiditis, sigmoid sinus thrombosis, and increased ICP treated by pediatric otolaryngology and neurosurgery services. We combined our patient's data with 104 children from 20 other reported series in the literature (Table 1). Data regarding age, gender, clinical presentation, imaging findings including affected dural venous sinus(es), surgical intervention, medical therapy, and the most recent clinical and radiographic follow-up were recorded from each series.

## Case Report

### *History and Physical Examination*

The patient is a previously healthy 6-year-old boy who presented to the emergency department with a 3-week history of progressive right-sided ear pain, headaches, nausea, and emesis. The child initially presented with symptoms of severe ear pain and fevers at an outside facility. His ear exam was reportedly normal, and he was prescribed azithromycin and amoxicillin/clavulanate. He followed-up with his pediatrician the following day, and was subsequently diagnosed with otitis externa and prescribed ciprofloxacin otic drops. His ear pain and fevers improved over the next several days; however, he began to develop projectile vomiting, headaches, and blurry vision. For example, his parents noted that he would close one eye while playing video games. They took him to a different local emergency department where he was diagnosed with resistant otitis media, and was prescribed cefdinir. He was instructed to follow-up with an optometrist urgently. His activity level remained normal, but his headaches, emesis, and visual symptoms continued. The family noticed that his eyes were starting to cross inward, and an evaluation by an optometrist revealed a diagnosis of papilledema.

On evaluation at our facility, the child was alert and interactive with no focal neurologic deficits other than bilateral abducens nerve palsies. A computed tomography of the head showed right-sided mastoiditis, with no intracranial abnormalities. ENT and neurology specialists were then consulted, and he also underwent a MRI/MRV (magnetic resonance imaging/magnetic resonance venography) that showed a right sigmoid, distal transverse, and partial internal jugular venous sinus thrombosis (see Figure 1). The left transverse sinus was hypoplastic with partial clot within it as well. A small volume lumbar puncture was performed with an opening pressure of 55 cm H<sub>2</sub>O. A mastoidectomy was recommended to debride the mastoid air cells, and

neurosurgery was consulted for the finding of the sigmoid sinus thrombosis and concerns for elevated ICP. As the child was awake, alert, and otherwise nonfocal despite the abducens nerve palsies, we recommended starting acetazolamide for ICP management, aggressive fluid hydration given his history of emesis, and starting a heparin drip as soon as possible once his mastoidectomy was performed. Hematology was also consulted for thrombogenic workup.

### *Operation*

A right complete mastoidectomy with myringotomy and tube placement was performed urgently by otolaryngology. A copious amount of purulent fluid was encountered once the mastoid was opened with a cutting bur, which was sent for microbiology. Granulation tissue was found to be adherent to the sigmoid sinus, as well as the mastoid antrum. The cavity was copiously irrigated and closed.

### *Pathology/Data*

On lumbar puncture, the spinal fluid was colorless and clear with no xanthochromia. The total neutrophil count was 0 and the differential was neutrophils 8%, lymphocytes 84%, and monocytes 8%. Glucose was 54 and protein was 16. Gram stain and the finalized culture were negative.

On serology, he had a normal basic metabolic profile, erythrocyte sedimentation rate, and C-reactive protein level, but an elevated D-dimer of 94. His hematologic workup included a complete blood count with differential that was within normal limits. He also underwent testing for hypercoagulable conditions that was also negative.

Microbiology tests were negative in his spinal fluid, blood, as well as his surgical cultures including the mastoid effusion and middle air fluid.

### *Postoperative Course*

The neurosurgical team reevaluated the patient immediately as he was emerging from anesthesia and verified that he was neurologically stable. By the next morning, the patient denied having any headaches and was watching TV without any visual complaints. His abducens nerve palsies were completely resolved. He was continued on oral linezolid and cefdinir empirically after his cultures remained negative and lovenox for anticoagulation for a total of 6 months. An MRI performed 6 months after his presentation showed complete resolution of his dural sinus venous thrombosis.

**Table 1.** Summary of Clinical and Radiographic Details of 105 Children, Including the Present Case, With Orogenic Dural Sinus Thrombosis in the Literature.

Author (Year)	Patient No.	Age (Years)	Gender	Chief Complaint	Neurologic Complications	Radiographic Findings (Including Affected Sinus)	Surgical Treatment	Medical Treatment	Length of Anticoagulation (Weeks)	Follow-up (Months)	Clinical Outcome	Radiographic Outcome
Scherer (2017)	1	7	1 male	Headache, otalgia, blurry vision, nausea/emesis	Papilledema, bilateral CN VI palsies	1—SS, TS, IJV	Mastoidectomy and myringotomy	Anticoagulation (heparin followed by LMWH) Antibiotics (linezolid and cefdinir) Acetazolamide	24	6	No sequelae	Complete recanalization
Ryan et al (2016) <sup>1</sup>	7	6-15	4males, 3 females	Fever, otalgia, otorrhea, nuchal rigidity	Papilledema	7 SS 7 TS 5 IJV 2 epidural abscess 1 HCP	7 mastoidectomy and tympanostomy tube 2 epidural abscess drainage	5 anticoagulation NOS 7 IV antibiotics	N/A	N/A	1—R VF defect 6—no sequelae	1 patient—improved flow but persistent thrombosis at 9 months
Zanoletti et al (2015) <sup>3</sup>	8	2-7	4 males, 4 females	Fever, headache, otalgia	6 papilledema 3 diplopia 1 torticollis 1 facial paralysis	8 SS 6 TS 4 IJV 1 epidural abscess	7 mastoidectomy 4 myringotomy	8 anticoagulation 6 acetazolamide 2 mannitol 8 IV antibiotics	12	8-27	No sequelae	6 recanalization 1 partial recanalization 1 persistent disruption
Singh et al (2014) <sup>28</sup>	6	10-25	4 males, 2 females	Fever, headache, otalgia, nausea, dizziness	N/A	6 SS 2 IJV	6 mastoidectomy 6 tympanoplasty 6 sinus needle aspiration	6 IV antibiotics 0 anticoagulation	N/A	3	No sequelae	N/A
Rosdy et al (2014) <sup>29</sup>	10	4-8	6 males, 4 females	Fever, headache, otalgia, otorrhea, lethargy, vomiting	9 papilledema 2 abducens nerve 5 torticollis 3 ataxia	10 SS 5 TS 3 IJV	10 mastoidectomy 2 open thrombectomy 1 IJ ligation 1 epidural abscess drainage	10 IV antibiotics 8 anticoagulation 9 acetazolamide	12-24	12	1 patient with unilateral visual deficit of 0.5 D	Complete recanalization in 50%, partial in 20%
Funamura et al (2014) <sup>30</sup>	5	1-15	4 males, 1 females	Fever, otalgia, headache, nausea/vomiting, lethargy, otorrhea	1 seizures after ICH	5 SS 4 TS 2 IJ	4 mastoidectomy 1 myringotomy tube 3 epidural abscess drainage	3 anticoagulation	12-24	1-5	1 seizures 1 headache	2 complete recanalization 2 partial 1 N/A
Ropposch et al (2012) <sup>5</sup>	6	3-15	6 males	Fever, headache, otalgia, nuchal rigidity, dizziness	1 abducens nerve palsy 1 papilledema	6 SS 3 TS 3 IJV	5 mastoidectomy 3 open thrombectomy 1 ligation of IJV	6 anticoagulation	12	3	No sequelae	6 recanalization

(continued)

Table 1. (continued)

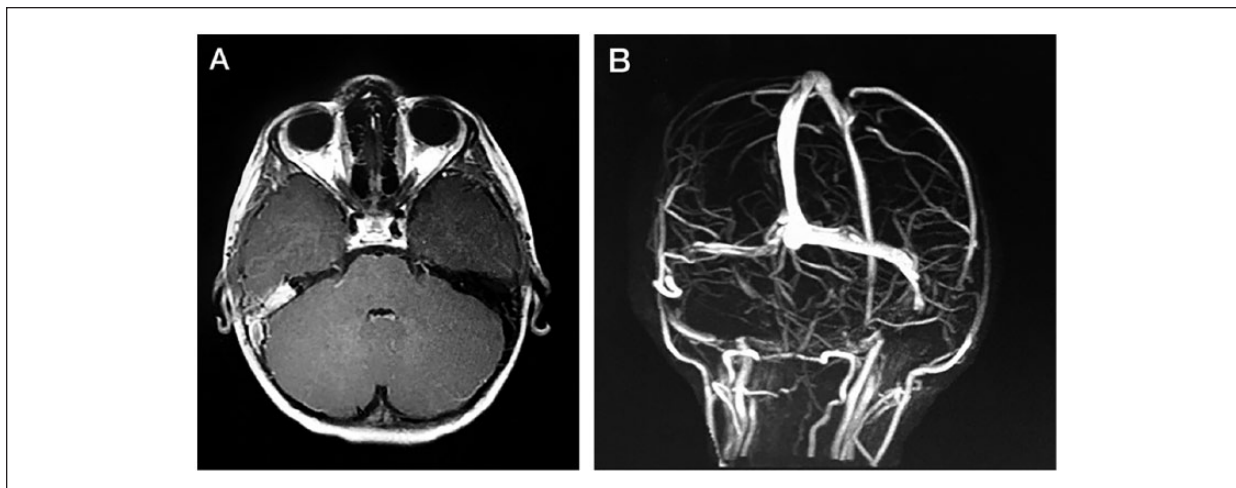
Author (Year)	Patient No.	Age (Years)	Gender	Chief Complaint	Neurologic Complications	Radiographic Findings (Including Affected Sinus)	Surgical Treatment	Medical Treatment	Length of Anticoagulation (Weeks)	Follow-up (Months)	Clinical Outcome	Radiographic Outcome
Sitton and Chun (2012) <sup>2</sup>	7	2-15	5 males, 2 females	Fever, otorrhea, vomiting, diplopia, otalgia, headache, mastoid tenderness, neck stiffness, dizziness	2 Papilledema	7 SS 3 TS 2 IJV	3 mastoidectomy with myringotomy 1 mastoidectomy and sinus aspiration 1 myringotomy and drainage of subperiosteal abscess 2 myringotomy alone 5 mastoidectomy	6 anticoagulation 7 IV antibiotics	24	1-7	No sequelae 1 epistaxis from anticoagulation that resolved with pressure	6 recanalization 1 thrombosis to IJV
Christensen et al (2009) <sup>16</sup>	7	4-12	5 males, 2 females	Headache, otalgia, nausea, CN palsies	2 papilledema 4 CN abducens nerve palsies 1 visual field cut 1 hearing loss	7 SS		6 IV antibiotics 1 oral antibiotics 1 anticoagulation (enoxaparin/warfarin)	1-2	1-4	1 lateral gaze diplopia 1 severe high-frequency hearing loss 1 field cut 1 persistent papilledema	4/6 showed recanalization 1 no imaging
Tov et al (2008) <sup>17</sup>	1	5	1 male	Otalgia, postauricular swelling	1 mild papilledema	1 SS	1 myringotomy	1 IV antibiotics	N/A	4	No sequelae	Recanalization
Saha et al (2007) <sup>18</sup>	1	11	1 male	Pyrexia, otorrhea, torticollis	1 Torticollis	1 SS 1 TS 1 IJV	1 radical mastoidectomy and open thrombectomy	1 IV antibiotics	N/A	NA	No sequelae	NA
Shah et al (2007) <sup>20</sup>	2	5-14	2 males	Fevers, otalgia, headaches, AMS	1 papilledema 2 abducens nerve palsies	2 SS	2 mastoidectomy 1 tympanostomy tube 1 sinus needle aspiration	2 IV antibiotics 2 anticoagulation	12-24	3-6	No sequelae	2 recanalization
Manolidis and Kurtz (2005) <sup>4</sup>	12	1-72	N/A	Fevers, otalgia, nuchal rigidity, CN palsies	1 papilledema 2 abducens nerve palsies 1 CN VII, IX, X, XI palsies	12 SS 4 TS 3 IJV 3 epidural abscesses 1 temporal lobe abscess 1 HCP	6 SS resection 6 open thrombectomy 1 VPS 4 mastoidectomy 1 subtotal temporal bone resection 6 canal wall up mastoidectomy 1 canal wall up mastoidectomy with ossicular reconstruction	12 IV antibiotics	N/A	N/A	No mortality	N/A

(continued)

**Table 1. (continued)**

Author (Year)	Patient No.	Age (Years)	Gender	Chief Complaint	Neurologic Complications	Radiographic Findings (Including Affected Sinus)	Surgical Treatment	Medical Treatment	Length of Anticoagulation (Weeks)	Follow-up (Months)	Clinical Outcome	Radiographic Outcome
Wong et al (2005) <sup>23</sup>	5	2-14	4 males, 1 female	Fevers, headache, otalgia, diplopia	1 abducens nerve palsy	5 SS 2 TS	2 mastoidectomy 2 myringotomy 1 therapeutic lumbar puncture	5 IV antibiotics	None	1-15	No sequelae	1 no recanalization 1 progressive recanalization 1 normal flow 6 doing well 5 N/A
Seven et al (2004) <sup>22</sup>	11	7-42	6 males, 5 females	Fevers, headache, otalgia	2 papilledema 4 AMS	11 SS	11 mastoidectomy 7 open thrombectomy 4 needle aspiration of sinus 2 epidural abscess evacuation	N/A	N/A	N/A	1 abducens nerve palsy 1 deaf 1 ataxia	
Agarwal et al (2003) <sup>24</sup>	1	3	1 female	Fevers, headache, otalgia	None	1 SS	1 mastoidectomy, posterior fossa craniectomy with epidural abscess evacuation, myringotomy tubes	1 IV antibiotics	None	1.5	No sequelae	Recanalization
Ooi et al (2003) <sup>25</sup>	4	13-23	1 male, 3 females	Fevers, headache, otalgia, diplopia	1 papilledema 1 abducens nerve palsy	4 SS	4 mastoidectomy 2 thrombectomy 1 sinus needle aspiration	4 IV antibiotics 1 anticoagulation	N/A	24 in the 2/4 who followed- up	No sequelae, but limited by follow-up	N/A
Bradley et al (2002) <sup>19</sup>	9	N/A	N/A	Fevers, headache, otalgia, AMS, otorrhea	6 AMS	9 SS 1 cavernous sinus 3 JIV	8 tympanostomy tubes 6 canal wall intact mastoidectomy 1 canal wall down mastoidectomy 1 needle aspiration of sinus 2 incision of sinus 2 VPS	9 IV antibiotics 6 anticoagulation	4-24	9	1 headache and otalgia	4/9 No evidence of thrombus progression
Spandow et al (2000) <sup>27</sup>	1	14	1 male	Fevers, headache, otalgia	1 papilledema	1 SS, TS, JIV	1 tympanostomy	1 IV antibiotics 1 anticoagulation	8	3.5	No sequelae	1 recanalization
Garcia et al (1995) <sup>26</sup>	1	7	1 male	Fevers, headache, otalgia, otorrhea	1 decreased hearing	1 SS, JIV	1 tympanostomy tube	1 IV antibiotics 1 anticoagulation	24	24	No sequelae	1 recanalization

Abbreviations: N/A, not available; HCP, hydrocephalus; NOS, not otherwise specified; VF, visual field; SS, sigmoid sinus; TS, transverse sinus; JIV, internal jugular vein; VPS, ventriculoperitoneal shunt.



**Figure 1.** (A) T1-weighted axial MRI with contrast shows abnormal contrast enhancement of the right mastoid portion of the temporal bone. (B) MRV demonstrates occlusion of the right jugular vein and sigmoid and distal transverse sinuses and left distal transverse sinus.

## Results

We reviewed a case of pediatric otogenic dural sinus thrombosis that was treated at our institution. This patient was combined with 105 cases from other series reported in the literature. Patient age ranged from 1 to 72 years. The male-to-female ratio was 2:1 (sex was not available in 21 patients).

Most patients presented with headache, fever, and otalgia. The most common findings on neurological examination were papilledema, cranial nerve palsies (abducens nerve palsy being the most common), and hearing loss. The sigmoid sinus was the most frequently affected dural venous sinus. All 105 patients underwent combined surgical and medical treatment, which included mastoidectomy (88 patients), myringotomy/tympanostomy (39), sinus aspiration (14), open thrombectomy (21 patients), internal jugular vein ligation (2), epidural abscess evacuation (10), ventriculoperitoneal shunt placement (4), serial LPs (2), ICP monitor (0 patients), and external ventricular drain (0 patients). At least 49 patients were treated with anticoagulation, but details were not provided on most of these patients on the selected agents. As for our patient, he was bridged on heparin and then placed on low-molecular-weight heparin, while another patient was treated with low-molecular-weight heparin and then warfarin. No patients were treated with aspirin. At least 16 patients ( $\geq 47\%$ ) were treated with acetazolamide, and 2 (2%) received mannitol. Antibiotics were given to at least 65 patients ( $\geq 62\%$ ). The mean follow-up was for 6 months (range = 1-27 months), during which at least 93 patients ( $\geq 89\%$ ) returned to their neurological baseline without deficits.

The most common deficits were visual field defects (3), headaches (2), hearing loss (2), and seizures (1). At least 46 patients ( $\geq 44\%$ ) of those who had follow-up demonstrated complete resolution of the dural sinus thrombosis on follow-up imaging studies; these studies were predominantly MRV but also included computed tomography venography (CTV) and angiography.

## Discussion

The anatomic location of the sigmoid sinus in the mastoid bone makes it vulnerable to the effects of the spread of infection from the middle ear. These effects include thrombophlebitis with impaired venous and cerebrospinal fluid (CSF) drainage from the brain resulting in increased ICP. The therapeutic dilemma for the pediatric neurosurgeon is how aggressive ICP needs to be managed especially while the patient undergoes general anesthesia for surgery with otolaryngology, and in the postoperative setting.

## Presentation

There was an age range of patients from 13 months to 72 years; however, children younger than 18 years were the predominant age group. The male-to-female ratio was 2:1. Most children presented with headaches, fever, and otalgia. At least 29 patients ( $\geq 28\%$ ) presented with papilledema, and 17 patients (16%) with cranial nerve VI palsies on neurological examination, reflecting increased ICP. Of the case reports in our review, most patients demonstrated small- to normal-sized ventricles on imaging, although 4 required ventriculoperitoneal

shunt placement from otitic hydrocephalus. It was not clear from the reports whether or not these 4 patients had developed ventriculomegaly. The presentation is most consistent with benign intracranial hypertension or pseudotumor cerebri.<sup>11</sup>

The most common sinus affected was the sigmoid sinus (100%), which is consistent with our understanding of anatomy and the pathophysiology of spread of infection.

## Treatment

**Conservative ICP Management.** Ninety-nine patients (94%) were treated without surgical intervention (eg, ICP monitor, external ventricular drain, serial lumbar punctures, and CSF shunt) for ICP management, albeit only 46 patients (44%) had papilledema or cranial nerve palsies indicating elevated ICP. The key step in management for these patients is the reestablishment of flow through the major dural venous sinuses. It is managed in most cases with systemic anticoagulation through the use of heparin to resolve the thrombus. Patients who demonstrate clinical deterioration while on heparin are often treated with endovascular strategies to recanalize the sinuses.<sup>9-15,21</sup> However, of note, 12 patients (11%) underwent open thrombectomy; none of the patients in our aggregate series necessitated endovascular thrombectomy. Children seem to respond very well to the course of antithrombotic therapy. We used acetazolamide as a temporary measure to control ICP in our patient; however, the routine use of acetazolamide in the management of ICP in this setting has not been borne out in the literature. Perhaps, our patient would have achieved a good outcome even without administration of acetazolamide.

**Surgical ICP Management.** On the other hand, none of the patients were treated with neurosurgical intervention. The tolerance to high ICP in children likely reflects the chronicity and gradual rise of the ICP. As opposed to an abrupt rise in ICP, a slow rise allows the compliance of the brain parenchyma, and intracranial blood and CSF volumes to adapt.<sup>11</sup>

## Clinical and Radiographic Outcomes

As it is in idiopathic and secondary forms of intracranial hypertension, the most feared severe and permanent complication of chronically elevated ICP is progressive blindness from post-papilledema optic atrophy. The average follow-up in our aggregate series of patients was 6 months (range = 1-27 months). Three of the reported patients demonstrated impaired visual acuity as a result of the transient rise in ICP from dural venous sinus thrombosis; however, observing these patients for

even longer periods could help determine whether the remainder of patients in our series are at a lifelong risk of developing late symptoms of visual deterioration. In addition to otolaryngology and neurosurgery follow-up, close ophthalmology follow-up is mandatory.

MRV was used in the majority of cases to evaluate the resolution of dural sinus thrombosis and help guide the duration of anticoagulation therapy. In 46 cases (44%), there was a documented resolution of the dural venous thrombosis with complete recanalization of venous outflow; improved but incomplete recanalization was documented in 17 patients (16%). The selection of MRV over CTV likely reflects a pediatric practitioner's reluctance in repeatedly exposing a young child to the effects of radiation from CT.

It is impossible to form strong recommendations with respect to the efficacy of conservative treatment versus surgery for ICP management in these complex rare cases based on the low level of evidence in the literature. However, based on the satisfactory results of conservative treatment in our single case and in our review of the literature of similar cases, it remains a viable option for neurologically stable patients without contraindications for full anticoagulation. We recommend conservative management with appropriate antibiotic and anticoagulation, with continued radiographic and clinical observation for children who do not present moribund, in extremis, or without neurological deterioration on therapy.

## Conclusions

From our experience and review of the literature, we recommend that pediatric patients with otogenic dural sinus thrombosis, even with increased ICP, be treated conservatively. Good outcomes were attained with mastoidectomy by otolaryngology without more aggressive surgical intervention for increased ICP by neurosurgery. The mainstays of treatment were appropriate antibiotics and anticoagulation therapy.

## Author Contributions

AS: Contributed to acquisition; drafted the manuscript; gave final approval; agrees to be accountable for all aspects of work ensuring integrity and accuracy.

AJ: Contributed to conception and design; contributed to analysis and interpretation; drafted the manuscript; critically revised; gave final approval; agrees to be accountable for all aspects of work ensuring integrity and accuracy.

## 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.

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