CASE REPORT DOI: 10.15713/ins.bhj.90

# A Case of Transversosigmoid Thrombophlebitis with Bilateral Subdural Empyema


## ABSTRACT

Subdural empyema is a life-threatening infection of the intracranial space, with very high rates of mortality if not appropriately diagnosed and treated. When managed well, with surgery and antibiotics, patients usually recover without significant residual deficits. The route of spread of infection and the organism involved needs to be investigated for complete eradication of the disease. We present the case of a young girl, without any comorbidities, initially diagnosed as having cerebral venous sinus thrombosis, and treated accordingly, who later suffered a cascade of complications that proved almost fatal. She was managed successfully at our institution, and recovered completely in the end, and is currently leading a normal life. A pathophysiological analysis of her disease process was made retrospectively and indicated how a seemingly innocuous dental infection spread over time to produce a life-threatening subdural empyema, involving a fastidious opportunistic commensal organism. We present an account of the same.


**Key words:** Aphasia, Empyema, *Streptococcus intermedius*, Subdural, Thrombophlebitis

## INTRODUCTION

Intracranial suppurative infections commonly spread either in contiguity from paranasal sinuses/middle ear or hematogenously from a distant source. Anaerobic and microaerophilic organisms play a major role in the causation of such abscesses. The organism in such cases may be difficult to isolate due to fastidious and exacting nature of requirements in culture media. The inability to obtain an antibiotic sensitivity report may complicate and prolong the duration of treatment. In all cases, the source of infection must be traced and eliminated to prevent recurrence of infection. In cases where all these steps are successfully carried out in a timely fashion, morbidity and mortality outcomes may be unexpectedly good. We describe the following case to illustrate our experience in dealing with a complicated case caused by an elusive pathogen.

## CASE REPORT

A 14-year-old girl, without any known history of medical comorbidities, developed mild intermittent headache. Having occurred around the time of examinations, it was attributed to stress and managed conservatively, leading to partial resolution. Three days later, she developed intermittent fever, mild neck stiffness, and speech abnormalities – semantic paraphasias and anomia. She was admitted to a private hospital, and on investigation, found to have left transverse-sigmoid sinus thrombosis [Figure 1a-c]. An investigation was undertaken to identify the cause of thrombophilia in this young female patient, and she subsequently tested positive for ACLA (anti-cardiolipin antibody) and LA (lupus anticoagulant). In conjunction with an elevated ESR (erythrocyte sedimentation rate) and CRP (C-reactive protein), she was labelled as a case of ACLA positive cerebral venous sinus thrombosis (CVST) and managed with Tab. Acitrom (acenocumarol). Blood cultures were found to be positive for *Streptococcus intermedius* (pan sensitive) and treated with antibiotics (2nd gen cephalosporins). The patient was discharged after 15 days, after resolution of fever and altered mentation.

Fifteen days after discharge, the patient had a recurrence of anomia, associated with high-grade fever spikes (2–3/day). Imaging indicated a left fronto-temporo-parietal (FTP) subdural hematoma (SDH) 4 mm thick [Figure 1d and e]. This was managed conservatively, in hospice, with an alteration of anticoagulant regimen. She was discharged after 15 days, on T. Rivaroxaban. Headache persisted, associated with a few episodes of vomiting.

Two weeks later, the parents brought the girl to our hospital, with complaints of fever off and on, headache, vomiting, and post-auricular pain. On examination, she was irritable, with positive meningeal signs, equal pupils, papilledema,


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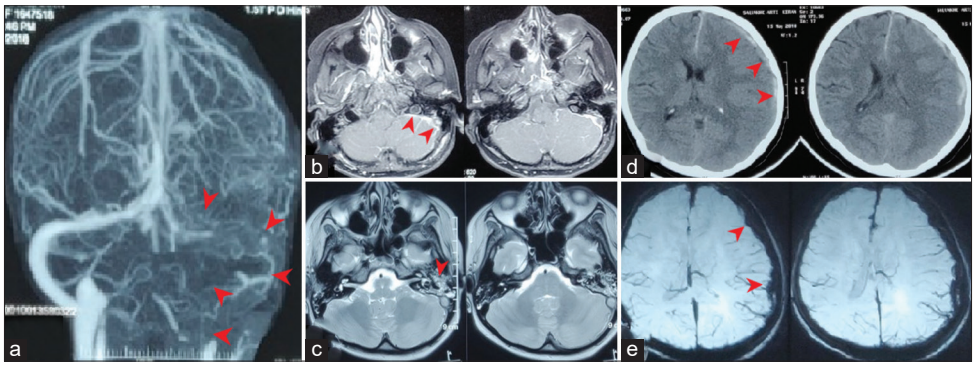


**Figure 1:** Primary imaging findings. (a) MRI contrast venography shows complete occlusion of the left transverse, sigmoid sinuses, and internal jugular vein. (b) Contrast-enhanced T1 MRI shows filling defects in the left transverse and sigmoid sinuses with adjacent dural enhancement. (c) T2-weighted MRI shows collection in the mastoid sinus and middle ear adjacent to the thrombosed sinus. (d) CT appearance of the left frontotemporoparietal subdural hematoma (SDH) as a complication of anticoagulant medication. E: MRI SWI showing left frontotemporoparietal SDH

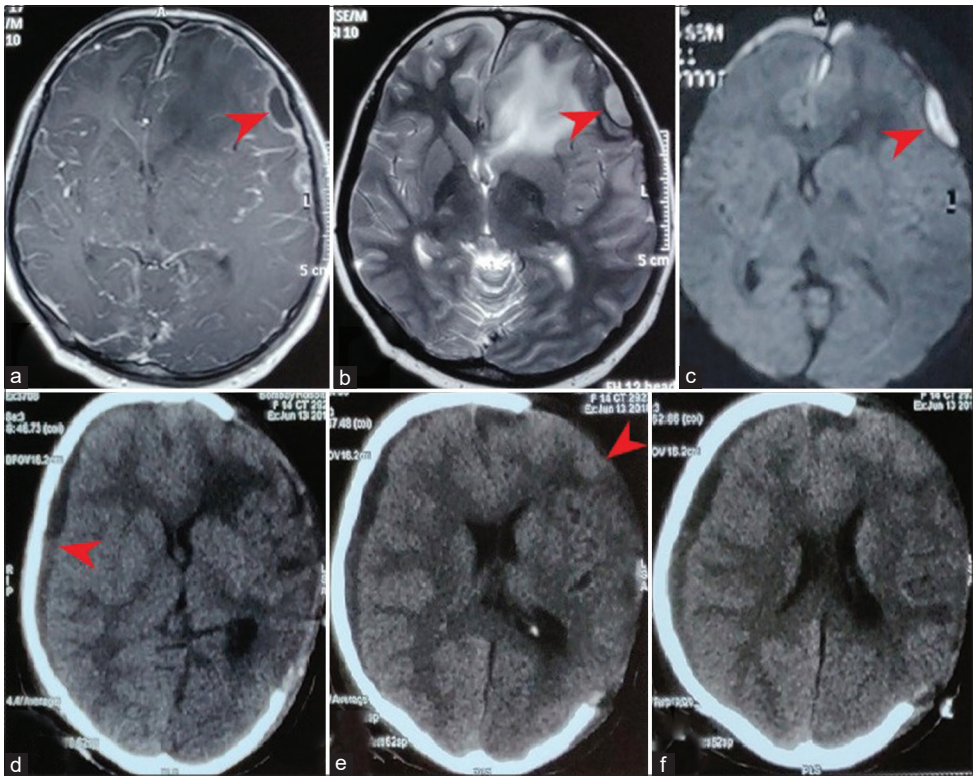


**Figure 2:** Progression of subdural hematoma to empyema. (a) Contrast-enhanced magnetic resonance imaging T1-weighted image showing left subdural peripherally enhancing collection. (b) T2-weighted image showing cystic collection. (c) DWI showing restriction, suggestive of empyema. (d-f) Post-operative CT scan showing right subdural collection with significant midline shift to the left and herniation of brain parenchyma through the craniotomy defect

mild right hemiparesis, right extensor plantar, and unsteady gait. Computed Tomography (CT) brain revealed a left FTP SDH, with a midline shift of 10 mm [Figure 2a-c]. She was taken up for immediate decompressive craniectomy and found to have a subdural empyema. Pus was sent for culture, the patient intubated and ventilated and started, on empirical antibiotic therapy. A search was undertaken to discover sources of infection in the body. However, the patient had a normal CT Abdomen, pelvis, and thorax. A 2D Echocardiogram was found to be normal. Sputum and urine cultures also turned out to be negative. The patient kept throwing fever spikes, while pus was found to be sterile. She underwent tracheostomy for airway management. One week later, fever settled down, the patient was awake, had anisocoria with Rt. pupil 3 mm dilated and sluggishly reactive, Glasgow Coma Score (GCS) $E_4M_5V_T$, persistent right hemiparesis (3/5), and poor comprehension.

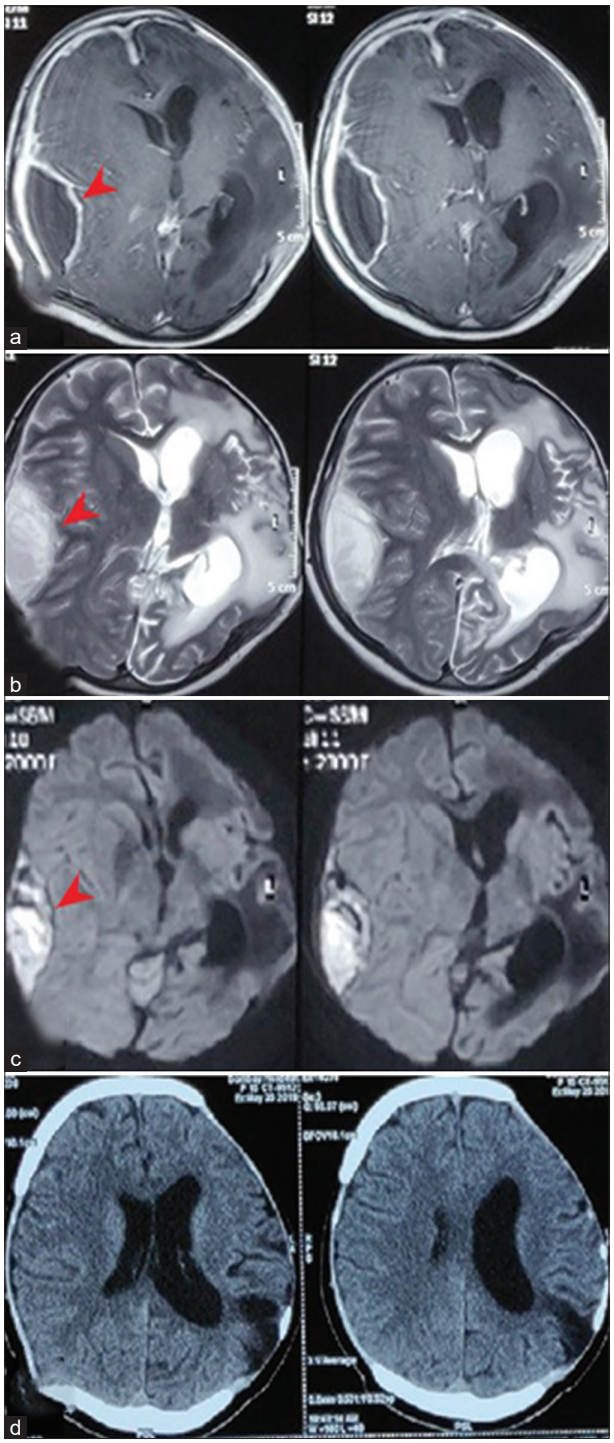


**Figure 3:** Development of contralateral subdural empyema. (a) Contrast-enhanced magnetic resonance imaging T1-weighted image showing peripherally enhancing collection in the right temporoparietal region with severe mass effect. (b) T2-weighted image showing cystic collection. (c) DWI showing restriction, suggestive of empyema. (d) Post-operative CT scan showing resolution of infective changes, with an area of gliosis in the left parietal region, in the setting of bilateral craniotomy defects

Imaging revealed a right FTP subdural collection (hematoma/empyema) [Figure 2d-f]. It was decided to observe her progress.

Two weeks later, the patient showed worsening of right hemiparesis (3/5 →2/5), recurrence of fever spikes, with a tense craniectomy flap. Contrast Enhanced Magnetic Resonance Imaging (CEMRI) revealed a large right FTP empyema [Figure 3a-c]. She was taken up for Rt. FTP decompression craniectomy and evacuation of empyema. The patient gradually improved and underwent decannulation of tracheostomy, and was subsequently discharged in the following clinical condition – right hemiparesis (3/5), global aphasia (motor>sensory), without any fever, and continuing physiotherapy. CT brain showed resolution of infective changes [Figure 3d].

Retrospectively, we found that she had undergone treatment for dental caries involving the left lower molar tooth 1 year before the first presentation, culminating in extraction of the tooth due to unresponsive suppurative infection (records not available). However, oropharyngeal examination was normal at the time of hospital admission.

At present (8 months later) – the patient has completely recovered, undergone bilateral autologous bone flap cranioplasty without adverse events, with no residual neuro/cognitive deficits, and is preparing to appear for her matriculate examination (Class X).

## DISCUSSION

*Streptococcus intermedius* is a Gram-positive microaerophilic bacterium belonging to the *Streptococcus milleri* group.[1] It is a normal commensal of the oral cavity, Gastrointestinal tract, and female Genitourinary tract.[1,2] It is the dominant organism isolated from dental plaque.[2] However, it displays remarkable opportunistic pathogenicity, even in immunocompetent hosts.[3] It produces numerous enzymes such as hyaluronidase, which allows it to disseminate its colonies throughout the body of the host by the way of bacteremia.[1,3] It has been reported to produce deep-seated suppurative infections, especially of the brain and liver,[1,3,4] as also infective endocarditis.[5]

The mode of spread of infection from its periodontal location may be contiguous, and hematogenous, or a combination of them.[6] This is usually precipitated by some manipulation of the tooth such as root canal, tooth extraction, or simple dental cleaning.[7,8] A thorough search must, therefore, be made in every case to locate the source of infection and treat it accordingly. The routes of spread of odontogenic infections are detailed in Figure 4.

In our case, the likely pathophysiologic mechanism is as follows:

Infection from the tooth spread through the pharynx, via the Eustachian tube to the middle ear and mastoid sinus. Here, it is likely to have spread into the jugular bulb, and produced sigmoid sinus thrombophlebitis. This spread retrogradely into the transverse sinus. It was misdiagnosed as a spontaneous CVST due to ACLA, and treated with anticoagulants alone. The patient developed bilateral subdural haematomas, left more than right,

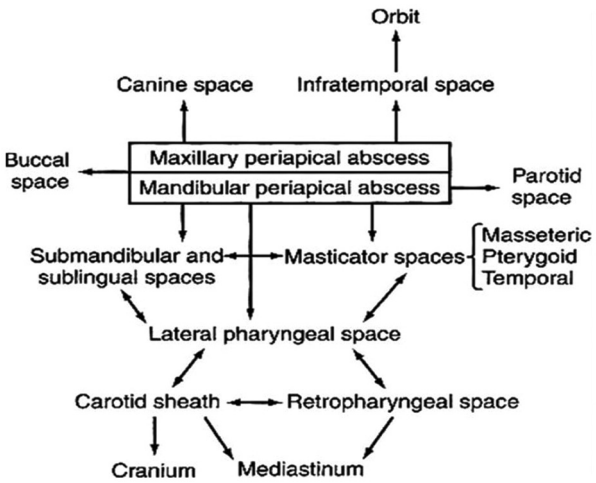


**Figure 4:** Flowchart showing the possible routes of spread of odontogenic infection through the deep fascial spaces of the head and neck

as a complication of anti-coagulant therapy. The haematomas were secondarily infected due to contiguous spread from infected transverse and sigmoid sinuses. The left-sided collection was evacuated first due to its larger size, and was then discovered to be an empyema rather than haematoma. The right empyema then expanded due to loss of tamponade effect due to the left decompressive craniectomy, and was subsequently evacuated.

Most isolates of streptococcus are sensitive to penicillin group of antibiotics; however, resistant strains are on the rise among *S. intermedius*. Such strains may be treated with vancomycin.[3,6] Empirical treatment may be started with the second-generation cephalosporins with anti-anaerobic agents. Most abscesses do not respond to treatment without drainage. Usually, stereotactic/percutaneous drainage is sufficient, since pus is sufficiently fluid, due to the presence of hyaluronidase.[3,4,6] However, in our case, decompressive craniectomy was necessary due to concomitant CVST with resulting cerebral edema and mass effect. Patients respond well with rarely any residual neurodeficit.[7]

The association of oropharyngeal sepsis with septic thrombophlebitis of internal jugular vein (IJV) is known as Lemierre syndrome. This is less commonly caused by *S. intermedius*.[9] Very few of these cases present with a CVST like picture, with subdural empyema.[10]

## CONCLUSIONS AND CLINICAL SIGNIFICANCE

- Anaerobic bacteria play an important role in the etiology of intracranial abscesses; hence, all pus samples must be subjected to aerobic, anaerobic, and capnophilic culture techniques
- *S. intermedius* is a commensal of the oral cavity, with a high degree of opportunistic pathogenicity. It is difficult to culture and isolate by routine microbiological techniques. It has a high propensity to produce deep-seated abscesses, especially of the brain, liver, and endocarditis. Spread occurs from periodontal foci through direct continuity or hematogenous routes
- Intracranial infections respond well to abscess drainage with antibiotic therapy. Cerebral abscesses usually resolve without residual neurodeficit
- A search must be made to find and eradicate source infections.

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**How to cite this article:** Shah SN, Bhagwat AA. A Case of Transverso-sigmoid Thrombophlebitis with Bilateral Subdural Empyema. Bombay Hosp J 2022;64(1):18-21.

**Source of support:** Nil, **Conflicts of interest:** None