

# Cryptogenic subdural empyema mimicking subacute-subdural hematoma on CT imaging in the pediatric group: a clinical case series

Mohammad Jaweed<sup>1</sup>, Dollicia Toh Ching Ying<sup>2</sup>, Esther Sim Wan Rong<sup>2</sup>, Mohammad Ajmal Yasin<sup>3</sup>, Azmin Kass bin Rosman<sup>2</sup>

<sup>1</sup>International Medical School, Management and Science University, Department of Neurosurgery Hospital Sungai Buloh, Malaysia

<sup>2</sup>School of Medicine, Faculty of Health and Medical Sciences, Taylor's University, Department of Neurosurgery Hospital Sungai Buloh, Selangor Malaysia

<sup>3</sup>Jamaica Hospital Medical Centre 8900 Van Wyck Expy, Richmond Hill, NY 11418, United States

To whom correspondence should be addressed: Dollicia Toh Ching Ying

e-mail: dollicia521@gmail.com

Available at:  
<http://www.archpedneurosurg.com.br/>

**Introduction:** A subdural empyema is a rare form of neurologic infection that can be attributed to a primary source of infection 60–70% of the time. Cases that cannot be attributed to a primary infection source are rare and deemed cryptogenic. Subdural empyema (SDE) is a rare but potentially life-threatening intracranial infection that is invariably fatal if left untreated. Clinical and radiologic features may be subtle because their presentation on neuroimaging may resemble subacute or chronic subdural hematoma. This report emphasizes the importance of high clinical suspicion, prompt diagnosis, and immediate surgical intervention, where craniotomy is preferred over burr-hole as a primary surgical option, given a favorable outcome in the treatment of SDE.

**Objective:** To explore the importance of early detection of subdural empyema based on high clinical and radiological suspicion, along with immediate surgical evacuation as a primary mode of treatment for SDE.

**Case presentation:** We present three cases of immunocompetent patients, 4 and 6-month-old infants with a 14-year-old adolescent presenting with SDE. The two infants underwent burr-holes as the pathology was initially diagnosed as a subacute-subdural hematoma, while intra-operatively found to be SDE. Subsequently, a wide craniotomy with intravenous antibiotics therapy was initiated. At the same time, the adolescent required a craniectomy because of the swollen brain.

**Conclusion:** Paediatric supratentorial SDE, although rapidly fatal if not identified promptly, can be effectively managed with early surgical drainage, preferably craniotomy, eradication of the source, and sensitive IV broad-spectrum antibiotics therapy.

**Keywords:** Subdural empyema, CT scan, Escherichia Coli, Infant, Acquired Community Infection

## INTRODUCTION

Subdural empyema (SDE), referring to intracranial purulent material found between the dura and arachnoid mater, are potentially life-threatening neurosurgical emergency if not treated properly(1, 2).

Anatomically, SDE is categorized as supratentorial, infratentorial, or affecting the spinal canal, of which the former forms the majority(1). In recent years, severe

neurologic sequelae and the mortality of subdural empyema have significantly diminished, with a survival rate of more than 90% given early detection, timely intervention, and adequate post-operative rehabilitation(1, 2). In the present reports, we retrospectively analyze our experiences with three cases of intracranial supratentorial empyema in the pediatric age group about aggressive surgical intervention and outcome.



## Cryptogenic Subdural Emyema Mimicking of Subacute-Subdural hematoma On CT Imaging in the Paediatric Group: A Clinical Case Serie

**Objective:** The objective of this paper is to shed light on the importance of early detection of subdural empyema based on high clinical and radiological suspicion, along with immediate surgical evacuation (craniotomy) as a primary mode of treatment for a better outcome.

### CASE PRESENTATION

We identified all patients in the pediatric age group (age < 14 years) with CT or operative evidence of SDE located in the supratentorial compartment through retrospective analysis of the case records between January 2019 to January 2021. Three patients were found to be qualified for our case report. All three patients were managed at Hospital Sungai Buloh, which is a tertiary hospital health facility in Malaysia.

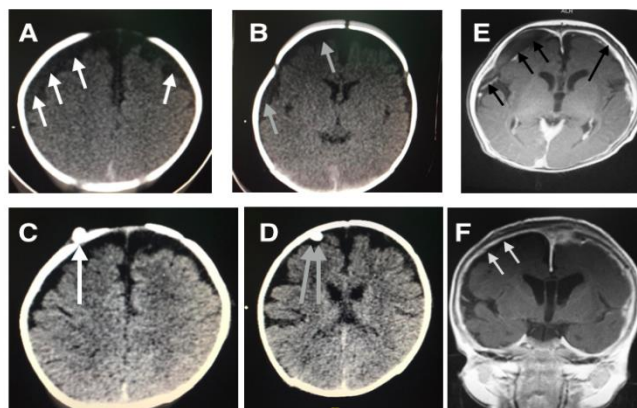
The electronic medical records, operation notes, and discharge summaries of all patients were analyzed retrospectively. Epidemiological, clinical, radiological, bacteriological, and outcome data were collected by a methodical analysis of the electronic case files of these patients. Predisposing medical conditions such as otitis media, paranasal sinusitis, hemophilia, congenital heart disease, and infective lung pathology were absent.

No apparent source could be identified in all 3 patients, and the parents denied any recent history of trauma or non-accidental shaking syndrome in the 2 infants, except for a urinary tract infection in the 6-month-old female infant. The subdural empyema was a sequela of meningitis in other 2 male patients (4 months and 14 years).

#### Case No. 1

A 4-month-old male infant came in with a high-grade fever for 2 days associated with photophobia. The infant developed 2 episodes of generalized tonic-clonic seizure on the second day of fever. The patient did not have any predisposing factors such as urinary tract infection, acute gastroenteritis, otitis media, head trauma, or upper respiratory infection, and he also did not have comorbidities such as congenital heart disease. The boy was born at term with normal vaginal delivery. His immunization was in line with the Malaysia National Immunization Schedule.

Upon physical examination, there was no specific abnormality except that he was febrile (temperature 39.1°C). Blood examination was normal except for a white cell count of 26.2 x 10<sup>9</sup>/L and a C-reactive protein level of 25.54 mg/L. Bacterial cultures of blood, urine, and stool were negative. Plain cranial computed tomography (Figure 1) revealed a bi-frontal temporal low-density subdural collection measuring 0.8cm and 1.0cm on the right and left,



**Figure 1-** Axial plain computed tomography scan and axial T1-weighted magnetic resonance images before (arrows A, B&E) and after (C, D&F) showing a low-density subdural fluid collection in the bilateral frontotemporal region (arrows A&B) of a four months old boy with mild midline shift. (C, D & F) showing postoperative subdural catheter in situ in the right subdural space (2 grey arrows-D). (F) administration of contrast medium performed eight weeks after completion of antibiotics demonstrating partial resolution of subdural collection (2 white arrows F)

respectively, with no midline shift or hydrocephalus. Basal cisterns and cerebral sulci were not effaced. He underwent bilateral burr-hole and subdural drainage. The subdural space was irrigated with a sterile saline solution. Cerebrospinal fluid examination revealed xanthochromia with protein 13.4g/L and gram-negative rods following the gram stain. The CSF culture grew *Escherichia coli*.

Postoperatively, the infant male became afebrile and was kept in the ward to complete his antibiotic therapy. Post-operative day 13, he developed 8 episodes of generalized tonic-clonic seizures. He underwent re-operative surgery and evacuation of empyema on the right side. He was discharged well after the completion of antibiotics, and on a recent follow-up, he is healthy with no mental or developmental delay.

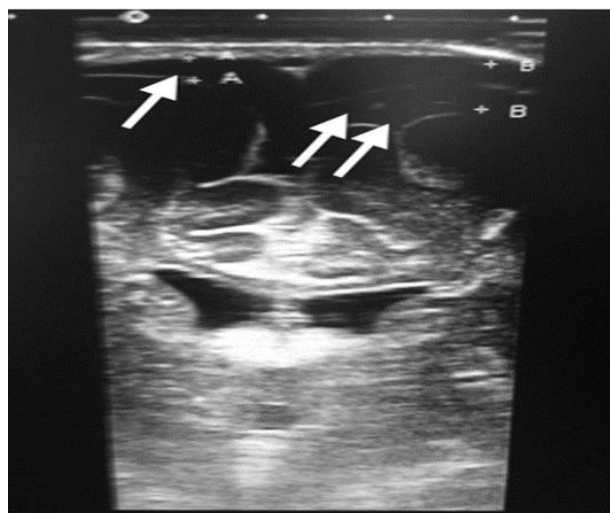
#### Case No. 2

A 6-months old girl born term came to us with status epilepticus. She had the previous neonatal jaundice admission and was subsequently well after discharge. Her illness started with one week history of high-grade fever. She appeared less active and developed vomiting on day 4 of the disease, followed by a focal seizure over the right upper limb that progressed into a generalized seizure. Upon physical examination, her fontanelle was full but soft; no specific abnormality was found except a ruptured bulla on her right shoulder. Her reflexes were brisk in all four limbs, attributed to a recent seizure. The parents denied recent head trauma or non-accidental injury. Blood examination disclosed peripheral white blood cell count 14.5 x 10<sup>9</sup>/L with neutrophilic predominance 86%, hemoglobin 8.2 g/dL, platelet count 95 x 10<sup>9</sup>/L, and C- reactive protein of 22.81 mg/dL. Urine analysis revealed traces of protein, but nitrate

## Cryptogenic Subdural Empyema Mimicking of Subacute-Subdural hematoma On CT Imaging in the Paediatric Group: A Clinical Case Serie

and leucocytes were negative. Urine culture was heavily mixed growth, most likely contaminant.

Chest radiography showed no abnormalities. Ultrasound of the Cranium shows bilateral Subdural collection confirmed by computed tomography, a low-density bilateral subdural collection in the front-temporoparietal region measuring 2.5cm and 2.0 cm on the right and left, respectively (Figure 2). Initially, she was planned for bilateral burr-hole surgery, which intraoperatively revealed turbid and foul-smelling fluid collection and converted to bilateral craniotomy, where the subdural space was irrigated with normal saline, and a bilateral subdural drain was inserted. Bacterial culture of purulent fluid isolated *Escherichia Coli*. However, blood and urine cultures were negative throughout admission. The infant was treated with a third-generation cephalosporin (cefotaxime 50mg/kg 6 hourly) for six weeks. Post-operative week 8, she developed communicating hydrocephalus, and a ventriculoperitoneal shunt was inserted. Her regular outpatient clinic follow-up was uneventful.



**Figure 2-** Cranial ultrasound showing bilateral subdural collection (arrows) measuring 0.7cm thickness on the right side and 0.9cm on the left side, respectively.

### Case No. 3

A 14-year-old male with no known medical illness was received from a peripheral hospital who presented with seizures and prolonged fever for two weeks associated with nausea, vomiting, slurring of speech, and neck stiffness for the one-day duration. Upon physical examination, his GCS was 15/15, and his left upper and lower limbs muscle power was 1/5 and 3/5, respectively. A diagnosis of right frontoparietal subdural empyema with mass effect secondary to meningoencephalitis was made (Figure 3). The patient underwent a craniotomy and evacuation of collection. During the operation, the patient developed

severe bronchospasm with carbon dioxide retention that led to a swollen brain that rendered us unable to put back the bone intraoperatively. Thus, the operation was converted to craniectomy with subdural washout. The subdural pus grew *Staphylococcus*; therefore, the patient was given IV antibiotics for four weeks. Post-operatively on day 6, non-contrast CT was done to reassess for subgaleal collection. The left cerebral sulci, ventricles, and basal cisterns were well preserved, and there was no midline shift. The patient was subsequently sent back to his original hospital. On the last follow-up, he was reported to be independent with no surgical complications.



**Figure 3-** A: Axial non-contrast CT image of 14 years boy showing a right-sided, frontotemporal, hypodense collection. B: Intra-operative image revealed the thick purulent yellowish collection underneath the dura, adherent to underlying arachnoid and brain parenchyma. C: Post-op axial non-contrast CT image showing right-sided decompressive craniectomy with a good evacuation of pus.

### DISCUSSION

SDE is the accumulation of purulent products between the dura mater and the underlying arachnoid mater(1, 2). Several aetiologies are found to be responsible for the formation of SDE in paediatric patients, the most common cause in infants being meningitis. Older children and adults are more predisposed to developing SDE in association with otitis media and paranasal sinusitis, the most common being frontal, followed by ethmoid, sphenoid, and maxillary sinusitis(3). Furthermore, iatrogenic causes such as lumbar puncture, subdural haematoma drainage, craniotomy, and intracranial pressure monitoring are known reported causes of SDE formation. Haematogenous spread causing SDE is rare, and the infective foci are most commonly from a pulmonary source or as a complication of osteomyelitis, trauma, or surgery(4, 5).

In our series, one infant (Case No.2) had haematogenous spread, as we believe the primary source of infection was the urinary tract, while the other two were idiopathic. SDE tends to be unilateral and supratentorial, as the falx and tentorium usually limit their spread to an area over one part of the hemisphere(1). However, with progression, there is a

## Cryptogenic Subdural Empyema Mimicking of Subacute-Subdural hematoma On CT Imaging in the Paediatric Group: A Clinical Case Serie

propensity for the empyema to spread rapidly throughout the subdural space and produce an expanding mass lesion effect with associated increased intracranial pressure and cerebral intraparenchymal penetration(6).

Causative agents of SDE are commonly bacterial in origin. These organisms include anaerobic and microaerophilic Streptococci and those in the Streptococcus milleri group, whereas Staphylococcus aureus, Escherichia coli and Bacteroides are other pathogens found. Pseudomonas aeruginosa or Staphylococcus epidermidis are said to be present in related iatrogenic cases, mainly pertaining to neurosurgical procedures. In addition, Salmonella spp. also plays a role in SDE formation in advanced AIDS patients(7). In our series, both patients 1 and 2 had Escherichia coli. However, in case 3, the causative agent was Staphylococcus. Clinically, a patient with SDE presents with a fever above 38°C, headache, a recent history of an SDE-predisposing factor (e.g., sinusitis, otitis media, meningitis, cranial surgery or trauma, pulmonary infection), meningism, altered mental status, hemiparesis, seizures, nausea and vomiting, speech or visual disturbances, and a rapid deterioration of consciousness(6).

All three patients in our series were alert, awake, and well-oriented, which are good prognostic indicators of surgical outcomes. The mortality rate is less than 10% for alert patients but can go up to 75% for patients comatose at the time of presentation(8). CT Brain is usually the first investigation performed and often is the only one required because patients, as a matter of course, proceed to surgery for intervention. The diagnosis of SDE is a challenge as its presentation on neuroimaging can greatly resemble that of subdural haematoma (SDH). Hence SDE requires high clinical suspicion. SDE typically appears crescentic in shape with a surrounding membrane that is markedly enhanced when contrast is introduced on imaging. The degree of rim enhancement seen in SDE is not normally found in cases of subdural haematoma, hence is a key feature in distinguishing SDE from SDH(9). MRI with gadolinium contrast has a higher sensitivity in detecting SDE and is also more effective in demonstrating SDE complications such as cerebritis, cerebral abscess, and venous thrombosis(1, 10, 11).

However, MRI machines may not be widely and easily accessible in the health care setting, which can further delay neurosurgical intervention(12). The treatment goal is to

reduce the ICP, evacuate the collection completely, and eradicate the source of infection. The advantages of craniotomy over burr-hole are evident as the former allows for broader exposure, adequate exploration, complete evacuation of the subdural purulent material, and decompression of the underlying cerebral hemisphere(13-15). Burr hole aspiration is sufficient when the pus is in thin liquid form, but as the disease progresses to thick pus, craniotomy is necessary to remove the larger quantity of infected material(12). In our series, 2 patients initially consented to bilateral burr-hole as early intervention given suspected SDH, but intraoperative findings converted the course of action to craniotomy because of SDE collection. Nevertheless, one patient (Case No.3) underwent craniectomy, given an intraoperatively bulging brain.

### DISCLOSURES

#### *Ethical approval*

This case series didn't require review by the Ethics committee from Hospital Sungai Buloh / Taylors University.

#### *Consent to publication*

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

#### *Conflict of interest*

The authors declare no conflicts of interest with respect to the content, authorship, and/or publication of this article.

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### CONTRIBUTIONS

Mohammad Jaweed: contributed to the study concept and design, writing and reviewing the paper.

Dollicia Toh Ching Ying: contributed to data collection and writing the paper.

Esther Sim Wan Rong: contributed to data collection and writing the paper.

## Cryptogenic Subdural Empyema Mimicking of Subacute-Subdural hematoma On CT Imaging in the Paediatric Group: A Clinical Case Serie

Mohammad Ajmal Yasin: contributed to reviewing the paper.

Azmin Kass bin Rosman: contributed to reviewing the paper.

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