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Novel Insights from Clinical Practice

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Interhemispheric and Infratentorial Subdural Empyema with Preseptal Cellulitis as Complications of Sinusitis: A Case Report

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Established Facts

- Subdural empyema is a rare case, accounting for less than 25% of total intracranial complications of sinusitis.
- The location, especially in infratentorial subdural empyema, is a challenge in diagnosis and treatment.

Novel Insights

- Prompt antibiotic selection is an essential therapy combined with neurosurgical intervention.

Keywords
Interhemispheric region · Infratentorial region · Subdural empyema · Preseptal cellulitis · Sinusitis

Abstract
Intracranial complications of paranasal sinusitis have become rare due to the use of antibiotics nowadays. However, several cases have been reported due the ability of paranasal sinusitis to cause serious complications. Once the infection spreads over the cranial structure, it could infect the orbits, underlying bones, meninges, adjacent veins, and brain. Subdural empyema is a rare but potentially life-threatening complication following paranasal sinusitis and should be considered as a neurological emergency. The location where subdural empyema may appear is a challenge in diagnosis and treatment. We report the case of a 17-year-old boy who presented in a state of somnolence due to interhemispheric and infratentorial subdural empyema with preseptal cellulitis secondary to pansinusitis. Early diagnosis and aggressive antibiotic treatment combined with neurosurgical operation were mandatorily implemented. The case was managed using a multidisciplinary approach including the ENT, eye, and nutrition departments. The boy achieved clinical improvement, with impairment of eye movement as the only persistent symptom before discharge. Daily supervision at the primary health care center with continuous antibiotic treatment was recommended to the patient. Pertinent literature with controversies in the management of subdural empyema will be briefly discussed in this case report.

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Introduction

Intracranial complications of paranasal sinusitis have become rare due to the use of antibiotics nowadays [1]. The intracranial complications of sinusitis are meningitis, intracranial abscess, subdural empyema, epidural abscess, cavernous sinus thrombosis, and thrombosis of other dural sinuses [2]. Subdural empyema is a rare but potentially life-threatening complication following paranasal sinusitis. It is defined as an intracranial focal collection of pus in a preformed space between the dura mater and the arachnoid mater [3]. Subdural empyema has been reported in 5–25% of intracranial infections [4]. The presenting symptoms sometimes appear not typical, so the diagnosis of such cases has been a challenge. Although rapid recognition and immediate neurosurgical intervention to evacuate pus together with appropriate antibiotic treatment give the patient a good chance of recovery with little or no neurological deficit, it is still a matter of controversy whether carrying out immediate neurosurgical intervention is as important as the early institution of appropriate conservative treatment [5]. Based on the location of pus, cases of interhemispheric and infratentorial subdural empyema have rarely been reported in the literature [6]. In this case report, we present the case of a 17-year-old boy with developing preseptal cellulitis as well as interhemispheric and infratentorial subdural empyema as the complications of sinusitis that was successfully managed using a multidisciplinary approach including neurosurgical evacuation and repeated antibiotic washout. Diagnostic and management strategies are discussed together with a review of the pertinent literature.

Case Report

A 17-year-old boy was referred from primary health care with a sudden decrease in consciousness, difficulties in speech, and paresis of the left side of his body. He had a history of allergic rhinitis with no history of head trauma. Firstly, he complained of malaise and fever for about 3 days, followed by headache on the left side of his head 2 days later. The symptoms worsened over 10 days with severe headache, fever, and vomiting, and 3 days prior to presentation, he also experienced swelling and pain in his left upper eyelid. When arriving at our emergency department, physical examination showed somnolence (Glasgow Coma Scale [GCS] score of E4, V4, M5); his vital signs were 160/90 mm Hg in blood pressure, a heart rate of 92 beats per minute, and a temperature of 38°C. His neck was rigid with an erythematous left eyelid, edema, and restricted movement of the left eye (Fig. 1a). Examination of his ears, oral cavity, and throat were unremarkable. His motoric functions of the left arm and the left leg were 2/5. In our emergency department, the patient experienced general seizure and was given 10 mg of intravenous diazepam.

Routine hematological examination revealed leukocytosis (23.5×10^9/L) with elevated segmented neutrophils (90.5%). C-reactive protein levels were grossly elevated (286 mg/L). An urgent noncontrast computed tomography (CT) of the head and paranasal sinuses showed interhemispheric subdural empyema which put pressure on the right ventricle and caused a midline shift to the left of 0.6 cm (Fig. 1b). The infratentorial area also showed hypodensity right under the cerebellar tentorium, suggesting infratentorial subdural empyema (Fig. 1c). There was near-complete opacification of the frontal and maxillary sinuses and diffuse bilateral mucosal thickening of all the sinuses, suggesting pansinusitis. There were also inflammatory changes within the left superior eyelid consistent with preseptal cellulitis.

After CT scanning, the patient was sent to the intensive care unit for further management, and neurosurgical consult was sought. He was initially managed conservatively with antibiotics. Intravenous ceftriaxone 2 g b.i.d., metronidazole 500 mg t.i.d., and gentamycin 160 mg q.d. were given as the main therapy with additional supportive medicine to lower intracranial pressure, such as 20% mannitol and 3% hypertonic saline boluses. Intravenous phenytoin 100 mg b.i.d. was also given to prevent seizure. Ophthalmology consultation was made to rule out orbital cellulitis, and surgical procedure to drain the pus from the palpebral edema was performed 1 day after admission. About a 3-cm horizontal incision was made at the upper eyelid under local anesthesia, and 3 mL of odorless yellowish-green pus was aspirated. The pus was sent for culture and showed bacterial growth of *Staphylococcus epidermidis*. The sensitivity result for the bacteria showed sensitive interpretations to amikacin, amoxyclav, amoxycillin, chloramphenicol, ciprofloxacin, ofloxacin, sulbactam cefoperazone, sulphamethoxazole, and teicoplanin. A diagnosis of preseptal cellulitis was made because there was no sign of orbital involvement. There was impairment in extraocular movement of the left eye (lateral movement) due to abducens (VI) nerve palsy. After 3 days of treatment in the intensive care unit, the patient regained consciousness and subfebrile temperature but still suffered headache, neck rigidity, weakness in extraocular movement, and left-sided hemiparesis. The patient was transferred to the ward with continuous conservative therapy. Seven days after admission, the patient underwent neurosurgical intervention using a craniotomy approach, and about 25 mL of white subdural pus was evacuated and drained. The subdural space was then repeatedly washed out with antibiotic solution (160 mg gentamycin in 10 mL normal saline). Pus was sent for culture but later failed to show any bacterial growth.

On the first postoperative day, the patient developed clinical improvements with no recurrence of headache, fever, or neck rigidity; motoric function was normal. On the sixth postoperative day, the patient was discharged from hospital with strict daily supervision at the primary health care center recommended for continuous antibiotics treatment. Intravenous antibiotics with gentamycin 160 mg t.i.d. were planned for the next 4 weeks. By the time of discharge, the only remaining symptom was restricted extraocular movement (lateral movement). The patient was advised to attend the ENT outpatient department for further management of his sinusitis. After 4 weeks of antibiotic therapy, the patient came to our outpatient clinic complaining of headache. There were no remarkable physical and laboratory findings. The patient underwent follow-up CT scan, and the results showed the remnant of the

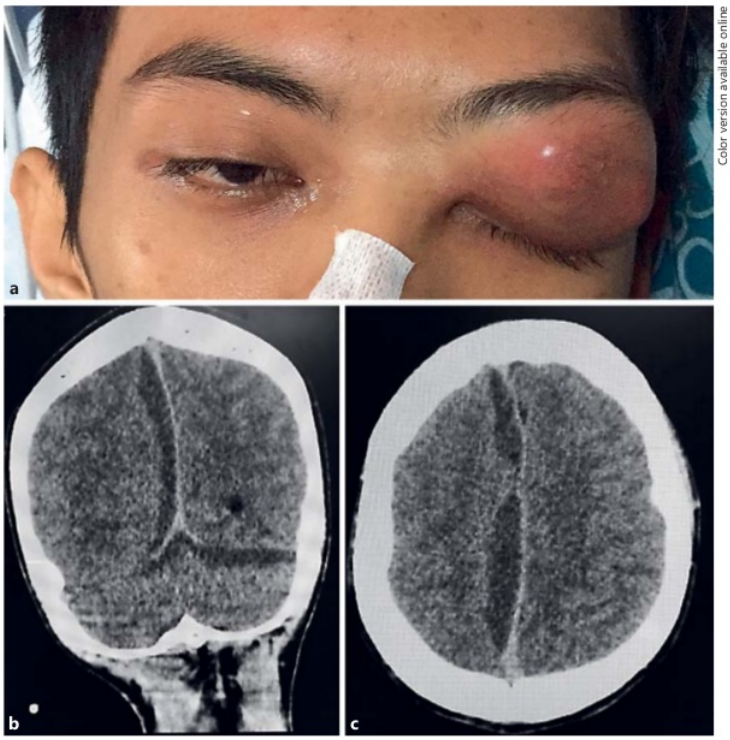

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Fig. 1. a Edema and erythema at left upper eyelid suggesting preseptal cellulitis. **b** Computed tomography scan with coronal view demonstrating infratentorial and interhemispheric subdural empyema. **c** Computed tomography scan with axial view demonstrating right interhemispheric subdural empyema.

previous subdural empyema (interhemispheric and infratentorial) with additional newly developed left frontal subdural hypodensity (Fig. 2). The patient refused to undergo further surgical intervention and asked for conservative therapy with antibiotic and symptomatic treatment only.

Discussion

Intracranial complications of sinusitis still occur and can be potentially life-threatening, despite the increasing use of antibiotics [1]. The intracranial complications of sinusitis are meningitis, intracranial abscess, subdural empyema, epidural abscess, cavernous sinus thrombosis, and thrombosis of other dural sinuses [2]. Subdural empyema has been reported in 5–25% of intracranial infections, with 80% of all cases presenting in males aged between 10 and 40 years [7, 5]. These findings are in accordance with our reported cases. However, the reason for this tendency of the majority of patients being young ad-

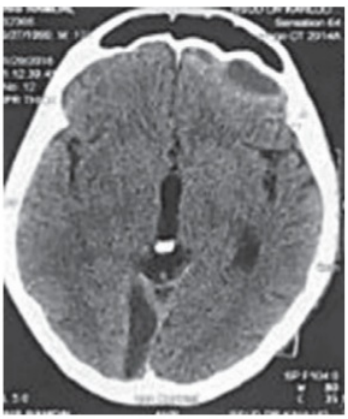

Fig. 2. Follow-up computed tomography scan with axial and coronal view demonstrating the remnant of the previous subdural empyema (interhemispheric and infratentorial) with additional, newly developed left frontal subdural hypodensity.

olescent males is still unknown [8]. The presentation of such intracranial complications is due to the spread of the organism from infected sinuses through septic thrombophlebitis of the emissary veins, which bridge the superficial cranial venous drainage and the intracranial venous system. Sometimes the organism spreads inward through Haversian channels in bone as a complication of osteomyelitis or skull defect, and it can also occur due to previous head trauma or neurosurgical procedures [9, 10]. Direct extension also plays an important role due to the close anatomic proximity of the sinus to the subdural space [11]. Once the infection reaches the subdural space from paranasal sinusitis, pus may extend further and can locate anywhere in the cranial subdural space [12]. In this case, our patient had a long history of untreated sinusitis. However, from the preformed CT scan, we did not find any skull defect or history of cranial operation. Therefore, the mechanism of the spread of infection to the subdural space is still unknown. We assume that the infection may spread via a hematogenous route or by direct infection through the haversian canal. In 1993, Bok and Peter [6] conducted a retrospective study of 90 cases with subdural empyema and found 34 and 4 cases, respectively, located at the interhemispheric (37%) and posterior fossa (4%), whereas the case series reported by Nathoo et al. [13] found that only 1 patient (0.5%) developed infratentorial empyema secondary to paranasal sinusitis. In 1999, Sahjpaul and Lee [14] also described a case of infratentorial subdural empyema secondary to paranasal sinusitis with pituitary abscess and septic cavernous sinus thrombophlebitis. Our patient suffered subdural empyema located in the interhemispheric and infratentorial regions. This combination is rare and we have not found any reported case involving 2 compartments in subdural empyema secondary to paranasal sinusitis, although there was 1 case reported by Kojima et al. [15] concerning supratentorial and infratentorial subdural empyema secondary to septicemia. In 2006, Venkatesh et al. [16] retrospectively analyzed 14 pediatric cases of infratentorial subdural empyema and found that in 3 patients (21.4%) it was associated with a supratentorial lesion. We assume that the pus originated in the supratentorial compartment and developed to infratentorial empyema by seeping through the edge of the tentorial incisura aided by gravity and changing head position [13].

The most common isolated organisms in subdural empyema are anaerobes, aerobic streptococci, staphylococci, *Haemophilus influenzae*, *Streptococcus pneumoniae*, and other gram-negative bacilli. At the same time, the most common organisms in subdural empyema secondary to paranasal sinusitis are anaerobic, microaerophilic streptococci, and in particular, those of the *Streptococcus milleri* group (*S. milleri* and *S. anginosus*) [7]. In this case, we found cultural bacterial growth of *S. epidermidis* from pus taken from preseptal cellulitis. *S. epidermidis* is a normal skin flora, but several cases have reported that it can cause pyogenic infection [17–19]; pus taken from the subdural space failed to show any bacterial growth. In some cases, it had been reported that pus culture showed no bacterial growth in 28.6% of patients [16].

Clinical presentations of subdural empyema are sometimes difficult to differentiate from meningitis with the most common presentation of subdural empyema being a triad of fever, sinusitis, and neurological deficits. The ensuing symptoms include nausea, vomiting, nasal symptoms, headache, seizure with no prior history, decrease in consciousness, history of intracerebral abscess, blurry vision, and speech difficulty (dysphasia). On physical examination, the signs of subdural empyema are changes in mental status, meningeal sign, hemiparesis or hemisensory deficits, aphasia or dysarthria, seizure, papilledema, homonymous hemianopsia, palsies of cranial nerves III, V, or VI, and signs of sinus inflammation such as swelling, tenderness, or redness [10]. In infratentorial subdural empyema, cerebellar signs have been reported to occur in 21.4% of patients. The signs include vertigo, ataxia, nystagmus, intentional tremor, slurred speech, hypotonic, exaggerated, broad-based gait, and disdiadochokinesia [16]. Because the signs and symptoms are almost similar to several differential diagnoses such as bacterial meningitis, brain abscess, epidural abscess, or cerebral thrombophlebitis, laboratory and imaging studies are needed [10].

Laboratory studies include white blood cell count, erythrocyte sedimentation rate, and C-reactive protein. These can be elevated and useful as early screening tools. Lumbar puncture is not recommended if there are signs of increased intracranial pressure. In subdural empyema, CSF finding from lumbar puncture may reveal an increased white blood cell count, an increased protein level greater than 100 mg/dL, and a decreased glucose level of 40 mg/dL; sometimes, the CSF is normal and does not show any bacterial growth [10]. We made a diagnosis of subdural empyema for this patient by full-blown signs and symptoms and by laboratory findings, which were supported by a noncontrast head CT scan. We did not do a lumbar puncture because it was contraindicated in this patient [10]. Suspicion of intracranial complications due to sinusitis was made by assuming the coexistence of orbital complications [20]. Singh et al. [21] reported extracranial complications such as preseptal or orbital celluli-

Table 1. Clinical profile, culture result, site of empyema, treatment, and outcome of reported cases of subdural empyema in pediatric patients

No.	Authors, year	Age, years/ sex	Clinical profile/physical findings/ laboratory findings	Empyema culture result	CT/MRI result	Antibiotic therapy	Surgical procedure	Outcome
1	Nicoli et al. [1], 2016	12/F	Clinical profile: headache, fever, vomiting, history of sinusitis Physical finding: a complete motor and partial receptive dysphasia after lumbar puncture Laboratory finding: CRP = 195 mg/L, WBC count of CSF = 111 E9/L	Micro-aerophilic *Streptococcus*	MRI showed left-sided maxillary and frontal sinusitis, left sided frontal epidural abscess (13×13 mm), 3-mm-thick subdural empyema around the left hemisphere, and wide leptomeningeal enhancement	Ceftriaxone, metronidazole	Beck's trepanation + maxillary antral washout and Burr hole + evacuation Subdural empyema and burr hole + evacuation of epidural abscess	Complete recovery
2	Bruner et al. [11], 2012	16/M	Clinical profile: fever, right temporal headache, witnessed tonic-clonic seizure, confused, left arm weakness, history of sinusitis Physical finding: 0/5 weakness in the left upper extremity and confusion to time and place Laboratory finding: WBC count = 17,300 cells/mm with a left shift	Not identified	CT showed right frontal subdural hypodensity; MRI showed right frontal sinusitis and a right subdural empyema with meningeal enhancement	Ceftriaxone, vancomycin	Method not mentioned	Complete recovery
3	Calik et al. [25], 2012	13/M	Clinical profile: frontally localized headache, decreased appetite, multiple cervical microlymphadenopathy, history of sinusitis Physical finding: N/A Laboratory finding: white blood cell count = 15 × 103/μL, CRP = 76 mg/L, ESR = 35 mm/h	Not identified	CT demonstrated mucosal thickening and a consolidated area in the left frontal sinus and in both maxillary sinuses; cranial MRI revealed frontal subdural empyema and a local edematous appearance suggestive of cerebritis in the brain parenchyma	Ceftriaxone, metronidazole	Craniotomy and drainage of the empyema + functional endoscopic sinus surgery and left frontal sinusotomy	Complete recovery
4	Patel et al. [8], 2016	10/F	Clinical profile: severe frontal headache, decreased appetite, fatigue, vomiting, seizure, history of sinusitis Physical finding: drowsy, posticta l state Laboratory finding: ESR = 95 mm/h, CRP = 1,2 mg/dL	Group A β-hemolytic *Streptococcus*	CT of the head and sinuses without contrast showed near-complete opacification of the frontal sinuses and diffuse bilateral mucosal thickening of all the sinuses; a 4-mm collection was seen in the anterior right frontal lobe of the brain subjacent to the right frontal sinus; MRI of the brain with and without contrast showed a right frontal sinus opacification and a 24 × 10.2 × 15.8 mm subdural empyema adjacent to the posterior aspect of the right frontal sinus	Vancomycin, linezolid, metronidazole	Craniotomy and drainage of the empyema + bilateral maxillary antrostomy + total ethmoidectomy + sphenoidotomy	Complete recovery
5	Bouziri et al. [24], 2011	7/F	Clinical profile: fever, vomiting, lethargic with cool, cyanotic extremities, history of poor oral hygiene Physical finding: neck stiffness (+), Kernig's sign (+) Laboratory finding: WBC count of CSF = 1,500 leukocytes (95% granulocytes), protein level = 0.7 g/L, glucose level of 0.4 g/L	*Streptococcus constellatus, Actinomyces viscosus*	CT showed a left hemispheric subdural empyema	Vancomycin, metronidazole, ampicillin	Not mentioned	Patient died

tis in 37% of patients with subdural empyema. CT scan was our first-line choice because it was readily available; however, several studies found that MRI is more sensitive than CT scan and is considered as the gold standard [22].

A favorable clinical outcome depends on early and accurate diagnosis supported by prompt treatment, even though controversies still exist regarding the treatment of subdural empyema. Subdural empyema should be considered as a neurosurgical emergency, which means that aggressive treatment including multiple antibiotic administration and neurosurgery procedure should be carried out as soon as possible [11]. The principle of source control by the drainage of pus has to be mandatorily implemented, on the top of which craniotomy has been included as one of the favorable prognostic factors [7], although nonsurgical treatment (only with antibiotics) is still acceptable [23]. The main purpose of neurosurgical operation is to decrease the toxic and inflammatory process in the brain and

surrounding vessels and also to reduce the mass effect of the subdural pus. Moreover, pus isolated from the subdural space can be used for identifying organism and antibiotic sensitivity [6]. Several varieties of surgical approaches have been used, but craniotomy and burr holes remain the most frequently used methods [6, 5, 16]. Irrigation of the site of empyema with saline with or without diluted antibiotics is another option during surgery [12, 16]. In our case, we implemented treatment using antibiotics and neurosurgical operation, although we did not perform early surgery due to the good neurological condition of the patient, with a GCS score of 15 and rapid clinical improvement after the first administration of antibiotics. Antibiotic therapy is the primary therapy for subdural empyema, and empirical antibiotics should cover gram-positive, gram-negative, and anaerobic bacteria. Antibiotic selection should include the following: (1) nafcillin, oxacillin, or vancomycin, plus (2) a third-generation cephalosporin, plus (3) metronidazole. Various times have been cited in the literature for the length of time antibiotics should be given, but ideally, antibiotic therapy should be 4 weeks or more [10]. Subdural empyema is associated with high morbidity and mortality. As shown in this case of a sinogenic infection with subdural empyema and preseptal cellulitis, early awareness, diagnosis, and treatment are essential. It is important to recognize intracranial complications which require immediate medical and surgical intervention. Various reported cases of pediatric subdural empyema with clinical profile, site of empyema, treatment, and outcome are summarized in Table 1 [1, 8, 11, 24, 25].

Statement of Ethics

Patient consent was obtained regarding the usage of patient data, and the patient accepted the terms and regulations under the hospital ethics committee.

Disclosure Statement

The authors report no conflicts of interest concerning the materials or methods used in this study or the findings specified in this paper. No funding was received for this study.

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