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CASE REPORT
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Subdural empyema, brain abscess, and superior sagittal sinus venous thrombosis secondary to *Streptococcus anginosus*

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What is already known on this topic?

- *Rhinosinusitis, which is caused by Streptococcus anginosus group bacteria in children, has a worse prognosis and serious complications compared with other pathogens.*
- *The necessity of surgical intervention and permanent neurologic damage rate is high in intracranial complications secondary to Streptococcus anginosus group bacteria.*
- *There are cases of cavernous sinus vein thrombosis caused by Streptococcus anginosus group bacteria in the available literature.*

What this study adds on this topic?

- *The development of rhinosinusitis secondary to Streptococcus anginosus in a pediatric patient without immunodeficiency can be complicated by subdural empyema that may need decompressive craniectomy, brain abscess, and superior sagittal sinus vein thrombosis simultaneously.*
- *To the best of our knowledge, this is the first case report of superior sagittal sinus vein thrombosis secondary to Streptococcus anginosus group bacteria in a pediatric patient in the available literature.*
- *The patient, who had sinus vein thrombosis with a very poor prognosis accompanied by subdural empyema and brain abscess, returned to her usual state of health without neurologic sequelae.*

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ABSTRACT

Streptococcus anginosus can be frequently isolated from brain abscesses, but is a rare cause of the liver, lung, and deep tissue abscesses. In this report, we present a patient with subdural empyema, brain abscess, and superior sagittal cerebral venous thrombosis as complications of rhinosinusitis whose purulent empyema sample yielded *S. anginosus*. A 13-year-old female patient was referred to our pediatric intensive care unit with altered mental status, aphasia, and behavioral change. On a brain computed tomography scan, subdural empyema extending from the left frontal sinus to the frontal interhemispheric area and left hemispheric dura was detected. Intravenous ceftriaxone, vancomycin, and metronidazole treatments were started. Subdural empyema was surgically drained. Postoperative brain magnetic resonance venography imaging showed superior sagittal sinus thrombosis. Cultures obtained from purulent empyema sample revealed *S. anginosus*. On the third day of hospitalization, a brain computed tomography scan showed brain edema, especially in the left hemisphere and significantly increased subdural empyema that had been previously drained. She was reoperated and decompressive craniectomy was performed. On the fifth day, partial epileptic seizures occurred. Brain magnetic resonance imaging showed a brain abscess on the interhemispheric area. The magnetic resonance imaging findings of abscess formation improved on 30th day and she was discharged on the 45th day after the completion of antibiotic therapy.

Keywords: Brain abscess, child, *Streptococcus anginosus*, subdural empyema, venous sinus thrombosis

Introduction

Streptococcus anginosus group (SAG) is a subgroup of viridans streptococci. The subgroup is composed of three different bacteria, which are known as *S. anginosus, S. constellatus*, and *S. intermedius, S. anginosus* (also known as *Streptococcus milleri*) is a member of SAG bacteria, which was identified by Guthof in 1956 by isolating it from a dental abscess (1). It is a natural member of the oral cavity flora, gastrointestinal tract, and genitourinary system. The bacterium is frequently isolated from brain abscesses, and it can rarely be isolated from deep tissue, lung, and liver abscesses (2). It is reported that rhinosinusitis, which is caused by SAG bacteria, has a worse prognosis and could cause serious complications when compared with other pathogens in the pediatric age group (3).

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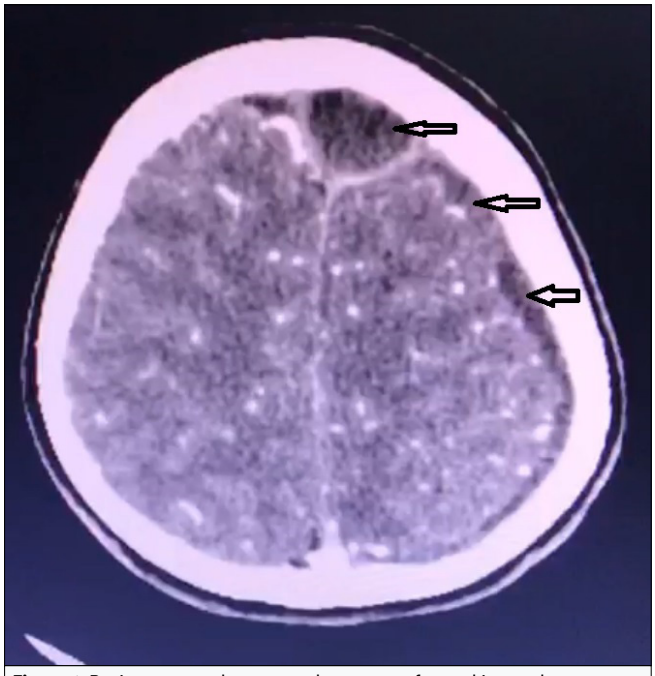

Figure 1. Brain computed tomography scan performed in another hospital. Subdural empyema extending from left frontal sinus to frontal interhemispheric area and left hemispheric dura (black arrows)

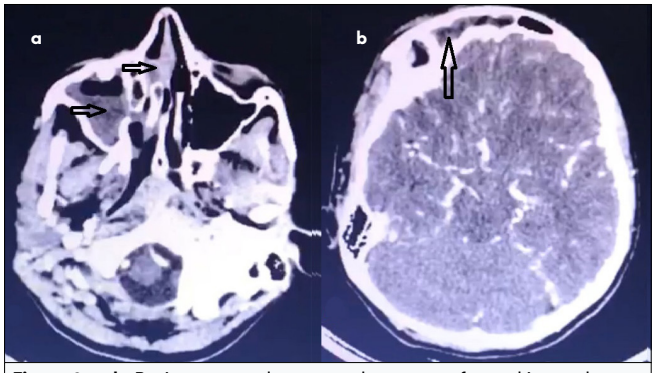

Figure 2. a, b. Brain computed tomography scan performed in another hospital. (a) Collection of dense fluid in compliance with rhinosinusitis in the right ethmoid and maxillary sinus (black arrows). (b) Collection of dense fluid in compliance with rhinosinusitis in the right frontal sinus (black arrow)

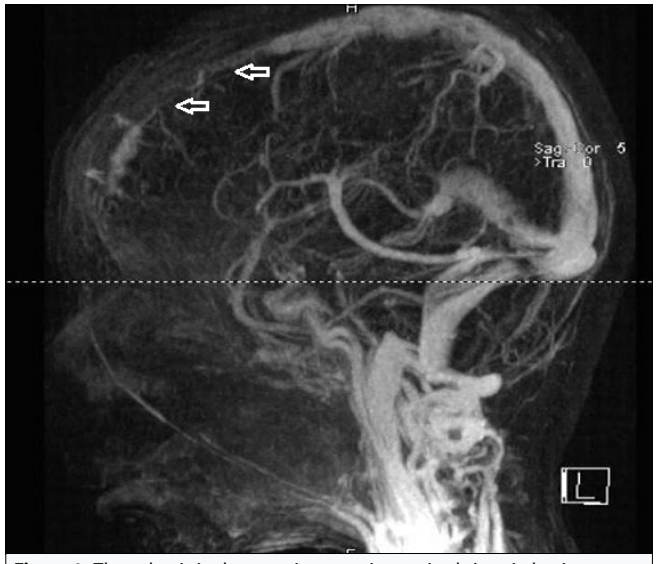

Figure 3. Thrombosis in the anterior superior sagittal sinus in brain magnetic resonance imaging venography scan (white arrows)

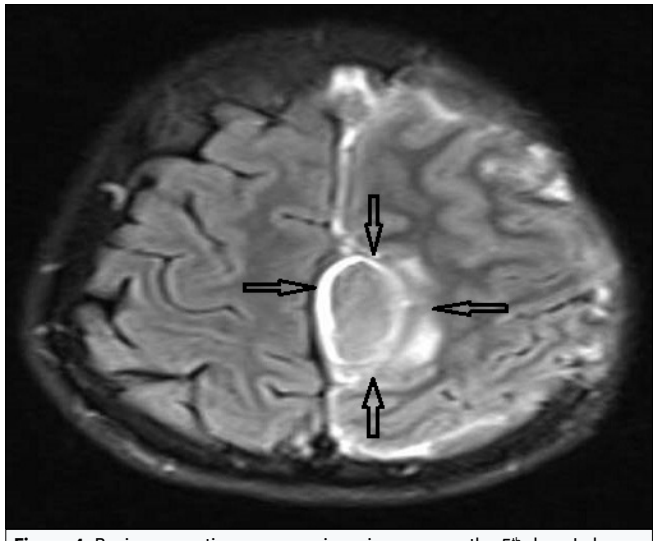

Figure 4. Brain magnetic resonance imaging scan on the 5th day. Lobulated and peripheral contrast enhancement brain abscesses in the interhemispheric area (black arrows)

Herein, we describe a case of a 13-year-old girl who had S. anginosus rhinosinusitis complicated by subdural empyema (SE), brain abscess (BA), and superior sagittal sinus vein thrombosis (SVT).

Case Presentations

A 13-year-old female patient was referred to our pediatric intensive care unit (PICU) with altered mental status, aphasia, bizarre behavior, and general muscle weakness. A physical examination in the PICU revealed somnolence (Glasgow Coma Score of 10), pupils were isochoric, and light reflex was present in both eyes. Heart rate, blood pressure, respiratory rate, and body temperature were normal. Respiratory effort was sufficient, lung auscultation was normal, and pulse oxygen saturation in room air was 96-98%. A complete blood count with differential revealed a white blood cell (WBC) level of 14 500/mm^3 (neutrophil level of 13 200/mm^3, lymphocyte level of 130/mm^3). The C-reactive protein level was 154 mg/L (normal; 0-5 mg/L) and the procalcitonin level was 4.24 ng/mL (normal <0.5 ng/mL). Other complete blood counts, biochemistry, electrolyte, and blood gas values were normal.

Subdural empyema extending from left frontal sinus to frontal interhemispheric area and left hemispheric dura was seen in a brain computed tomography (CT) scan that was performed in another hospital (Figure 1). Also, the brain CT detected a collection of dense fluid consistent with rhinosinusitis in the right frontal, ethmoid, and maxillary sinus (Figure 2). Her medical history was not significant for any trauma recently, she had no previous illness and had not been hospitalized previously. Before she was admitted into the PICU, the patient had headaches, fever, and postnasal drip for the last ten days, and was receiving oral amoxicillin-clavulanic acid treatment for her condition of rhinosinusitis.

Intravenous treatments of ceftriaxone, vancomycin, and metronidazole, which had been first started in the previous hospital, were continued. In order to prevent progression to possible brain edema, 3% sodium chloride (NaCl) infusion (1 mL/kg/hour) was initiated. Levetiracetam was also started for seizure prophylaxis. Empyema drainage was performed surgically. Severe cerebritis combined with decreased SE and thrombosis in the anterior superior sagittal sinus was noted in postoperative brain magnetic resonance imaging (MRI and a venography scan (Figure 3). Thus, enoxaparin was added to her current treatment. *S. anginosus* was detected in purulent and malodorous material, which was taken from subdural empyema, and it was susceptible to all antibiotics. However, her antibiotic regimen was not changed because of her poor clinical condition. The patient started to develop bradycardia and hypertension on the 3rd day of PICU admission. We decided to perform another brain CT examination due to a preliminary diagnosis of intracranial hypertension. The CT examination revealed notable brain edema, especially in the left-brain hemisphere and rapid increased SE, which was decompressed previously. Empyema drainage and decompressive craniectomy were performed. On the 5th day of PICU admission, the patient started to develop focal epileptic seizures, thus phenytoin was added to her current treatment. Repeated brain MRI showed lobulated and peripheral contrast enhancement brain abscesses in the interhemispheric area (Figure 4). Her current antibiotic regimen was changed to intravenous vancomycin and meropenem. Penicillin G was added to the treatment on the 6th day of PICU admission because the *S. anginosus* that grew in the empyema material culture was sensitive to penicillin (minimum inhibitory concentration <0.06). However, vancomycin and meropenem treatments were continued for 24 days due to her poor clinical condition, she underwent multiple brain surgeries including decompressive craniectomy to cover possible hospital infections and possible microorganisms that cannot be grown in culture. Frontal sinus drainage was performed due to possible leakage from the frontal sinus to the subdural space. The patient was lymphopenic throughout her PICU hospitalization. Her immunoglobulin G-A-M-E, lymphocyte subgroup examination, dihydrorhodamine test, and complement 50 levels were all normal.

On the 10th day of PICU admission, acute-phase markers regressed notably and body temperature became normal, and the patient became more conscious; therefore, she was transferred to a regular pediatric ward. On the 30th day of the hospital admission, another MRI of the brain was performed. The MRI revealed significant amelioration in findings of cerebritis and abscess; therefore, her antibiotics were stopped and intravenous ampicillin-sulbactam treatment was started. On the 45th day of the hospital admission, she was discharged from the hospital after completing the treatment of ampicillin-sulbactam for fifteen days.

During her one-year follow-up after she was discharged from hospital, she returned to her usual state of health in terms of consciousness, intelligence, and neurologic condition. Enoxaparin treatment was terminated. Her lymphocyte levels reached the normal range. An advanced genetic examination (whole-exome sequencing) was normal for immunodeficiency. Written informed consent was received from the parents of this patient for this case presentation.

Discussion

Before vaccines of Haemophilus influenzae and *Streptococcus pneumoniae* become prevalent, SE was more frequently seen in infancy. Nowadays, it is more frequently seen in childhood and young adulthood (4). Rhinosinusitis, especially in the frontal sinus, is the most prevalent underlying cause (5). Brain abscess is equally seen in both childhood and adulthood. The underlying causes of the brain abscess are as follows: immunodeficiency, infection in close body parts, previous traumas, brain surgery, and congenital heart diseases (6). Our patient was an adolescent and had a collection of dense fluid consistent with rhinosinusitis on the right frontal, ethmoid, and maxillary sinus, which was consistent with the literature. In our case, previous medical history, physical examination, and laboratory findings were not compatible with immunodeficiency and her advanced gene analyses resulted as normal.

S. anginosus bacteria are more prevalent and significant causes of rhinosinusitis in children and also have a more frequent complication rate than other bacteriologic agents (3, 6). Deutschmann et al. (3) analyzed 50 hospitalized and immunocompetent children with rhinosinusitis accompanying intracranial complications in their retrospective study. The result of their study was that the most common etiologic factor in these pediatric patients was SAG bacteria (14 patients, 28%), followed by *S. pneumoniae* (10 patients, 20%). The same study also reported that the necessity of surgical intervention and development of permanent neurologic damage secondary to intracranial complications were found to be higher in the SAG bacteria group (3). Cole et al. (6) investigated 42 pediatric patients with both SE and BA. The most prevalent etiologic factor was SAG bacteria with 30.95%. In compliance with the available literature, both rhinosinusitis and the complication of SE necessitated surgical intervention in our patient. The surgically drained SE increased in a short amount of time and constituted a severe threat to the patient's life due to the severe increment of the intracranial pressure. Therefore, we had to perform decompressive craniectomy. At the same time, the patient developed BA while she was under antibiotic treatment.

Cerebral SVT is one of the complications of acute or chronic sinusitis, and cavernous SVT is frequently encountered. The probability of the development of superior sagittal SVT is very low. The underlying cause of the pathophysiology is retrograde venous thrombophlebitis. Superior sagittal SVT has the worst prognosis, with a mortality rate of 80% (7). Even though there are a few case reports of cavernous SVT caused by SAG bacteria (8, 9), to best of our knowledge, this is the first report of SAG bacteria causing superior sagittal SVT in a pediatric patient in the available literature. The patient returned to her usual state of health without any neurologic sequelae, even though she had the worst prognosis of SVT combined with SE and BA.

In conclusion, rhinosinusitis, which was caused by *S. anginosus* and other SAG bacteria, has a worse prognosis compared with other pathogens. It should be kept in mind that it might also cause severe complications of SE, BA, and cerebral SVT.

Informed Consent: Written informed consent was obtained from patients' parents who participated in this case.

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Conflict of Interest: No conflict of interest was declared by the authors.

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