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Unusual case of Lemierre's Syndrome in a 5-year-old

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Abstract

Streptococcus pyogenes is a common cause of invasive infections. The role of Streptococcus pyogenes in the combination of meningitis and Lemierre's syndrome is rare.

A case of group a streptococcal meningitis is reported in a previously healthy 5-year-old boy. He presented to the emergency room with an altered level of consciousness; 16S ribosomal RNA test was positive for streptococcus pyogenes found by his spinal fluid, all cultures were negative. His course was complicated by a left-sided sinus vein thrombosis with extension to the external jugular vein. The extension to the neck diagnosed as Lemierre's syndrome.

Our case indicates that Streptococcus pyogenes infection should be suspected in case of Lemierre's syndrome combined with meningitis.

Abbreviations

CT: Computed Tomography; CSF: Cerebrospinal Fluid; IV: Intravenous; MRI: Magnetic Resonance Imaging; SVT: Sinus Vein Thrombosis.

Background

Streptococcus pyogenes is a rare pathogen of meningitis. However invasive infections as bacteraemia, pneumonia, skin and soft tissue, and toxic shock due to S pyogenes are common. Very few cases of Sinus vein thrombosis as a complication of S pyogenes have been reported [1]. Children suffer from associated complications much more than adults. This case report point out an uncommon presentation of meningitis due to S pyogenes [2].

Case Presentation

A 5-year -old boy arrived to the emergency room with deep unconsciousness. Over the last month prior admission, he experienced intermittent episodes of fever and headache. His immunizations were up to date including vaccine against Streptococcus Pneumoniae and Haemophilus Influenza.

He presented with moderate dehydration, temperature of 38.8°C and lethargy, with a Glasgow Coma Scale of 7/15. Nuchal rigidity was elicited. His eyes deviated to the left. The remainder of his physical examination was within normal limits. Opacification of the left mastoid documented with a Computed Tomography (CT) scan of the head. No obvious bony destruction was noted. A possible area of fluid or pus adjacent to the left sigmoid and transverse sinus was noted. There were no signs of thrombosis. Laboratory tests documented a white blood cell count of 14×109/L (93% neutrophils and 7% lymphocytes). The Cerebrospinal Fluid (CSF) cell count documented a nucleated cell count of 509×106/L, (72% neutrophils), protein level 0.6 g/L (normal 0.2 g/L to 0.4 g/L) and glucose level 0.2 mmol/L (2.2 mmol/L to 3.8 mmol/L). The CSF gram stain did not revealed any pathogen; Subsequently there was no growth of bacteria. 16S ribosomal RNA test of the spinal fluid was positive for streptococcus pyogenes. A peripheral blood culture had not grown any organism. Spinal fluid for acid-fast bacilli and fungi were negative. The patient was admitted to the pediatric intensive care unit, and was started on intravenous (IV) vancomycin (60 mg/kg/day) [3], IV meropenem (120 mg/kg/day) [4] and acyclovir (30mg/kg/day) [5]. Magnetic resonance imaging (MRI) of the head, showed diffuse abnormal sign in the left mastoid bone with contrast enhancement, left jugular vein thrombosis and left lateral Sinus Vein Thrombosis (SVT), and enhancement of the 7-8 left cranial nerves. Due to thrombosis visualized and abnormal coagulation studies, treatment with enoxaparin (2 mg/kg/d) was initiated. Metronidazole was added as empirical treatment for optional fusobacterium necrophorum infection until bacterial

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culture came back negative.

The patient began improving within 48 hours. His level of consciousness improved, and he could complain of left ear pain. Eye deviation improved, as well. On day 4 of his recovery, he was afebrile. Day 6 post admission, he started complaining of headache with vomiting once a day until the 8th day. On day 11, due to new-onset fever up to 38.5 Celsius degrees and deterioration in conscience levels, another MRI was performed because of concern of autoimmune encephalitis. The MRI revealed improvement of the mastoiditis, jugular vein thrombosis and sinus vein thrombosis and no signs of encephalitis. Subsequent LP demonstrated clear CSF with nucleated cell count of 417-60% PMN, protein level 73, glucose level 39.

Since clinical deterioration under treatment documented, autoimmune process was suspected. Therefore, autoimmune evaluation was performed. The evaluation included GABAB Ab, AMPAR1 Ab, AMPAR2 Ab, LGI1 Ab, CASPR2 Ab, NMDR Ab and Autoimmune Ab. The evaluation results did not support autoimmune encephalitis.

Finally, the patient was discharged after 21 days in the hospital. He was enrolled to a local home IV program to complete four weeks of IV ceftriaxone, which was changed to oral amoxicillin and also six weeks of oral metronidazole and 3 months of enoxaparin. Hematologic consultation after discharge didn't reveal any coagulation abnormalities including Factor V Leiden, Prothrombin genotype, Serum level of Protein C, Protein S and Antithrombin, APLA, factor 8 and D-DIMER.

IgG, IgM, CH-50, INR, PT, aPTT was in normal range, IgA was slightly low.

MRI 2 months after discharge and 5 months after discharge revealed irregularity of the left transverse vein and left sigmoid vein, raising the possibility of an old thrombus with partial recanalization.

Enoxaparin treatment was gradually tapered to once a day and after 6 months was stop.

Discussion and conclusions

A case of aseptic meningitis with Lemierre's syndrome is presented. Wide range of pathogens were considered as an optional agent, even M. tuberculosis and fusobacterium necrophorum. The final diagnosis was made based on positive results from the 16S ribosomal RNA test that identified Streptococcus Pyogenes [6-8].

The most unique aspects of this case arise from the fact that the initial presentation pointed towards meningitis and a suspected seizure, when in fact this was an atypical presentation for Lemierre's syndrome. The most common symptoms at presentation of Lemierre's syndrome include sore throat, trismus and neck pain. Lemierre's syndrome has only been described in association with meningitis in1 pediatric patients on two occasions both caused by Fusobacterium Necrophorum with one of them presenting with a cerebral infarct [9] and the second one with neurological and pulmonary involvement [10].

The most common bacterial culprit of Lemierre's syndrome is the anaerobic Gram-negative rod, Fusobacterium necrophorum identified mainly by blood cultures. Up to 10.1% of reported cases consisted of polymicrobial infections including Fusobacterium. About 5.5% of cases involved an organism other than Fusobacterium Necrophorum [11]. Most notably, however, 12.8% of cases grew negative cultures. In our case the pathogen that we found in the cerebral spinal fluid was facultative anaerobic GAS, while the blood cultures were negative. Only one other pediatric case of Lemierre's syndrome caused solely by GAS have been reported [12]. Furthermore, in case of Lemierre's syndrome, Streptococcus pyogenes had been implicated only a few times as a polymicrobial organism. Therefore, it is strongly suggesting its rarity as a cause of this disease [11].

In our case, the situation that all cultures were negative, until the 16s rRNA test resulted and identified Streptococcus pyogenes is important. A study of 394 samples from primary sterile body sites found that there was a high concordance of both culture and PCR (90.6%) and discordance in 4.6% with culture-negative, PCR-positive and 4.8% culture-positive, PCR-negative results [6]. Another study included 342 blood samples from 187 patients (173 adults and 14 children younger than 18 years), blood culture was positive in 54 samples (15.8%) from 34 patients. Of these 54 samples, 47 were also PCR positive. Among the 288-blood culture negative samples, a positive PCR result was obtained in 41[7]. In another study, 233 blood culture samples were examined by PCR-hybridization and compared with culture results. 123 from 128 positive culture contained a single bacterial species with 99.2% concordance by PCR-hybridization. 94% of the culture-negative samples, were also negative by PCR-hybridization. Most of the culture negative, PCR-hybridization-positive samples, contained CONS [8].

In this case report, we present a case of group A streptococcal meningitis and mastoiditis complicated by Lemierre's syndrome and left lateral sinus vein thrombosis in a previously healthy 5-year-old boy. In addition to the atypical presentation, the atypical bacterial culprit makes this essential bringing it to light. Only a few cases of meningitis and Lemierre's syndrome due to GAS have been reported previously in the literature. Our case support adding GAS to the list of differential causative agents of meningitis and Lemierre's syndrome. Moreover, it expands our understanding of the possible infectious etiologies that underly these conditions, specifically in the pediatric population.

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