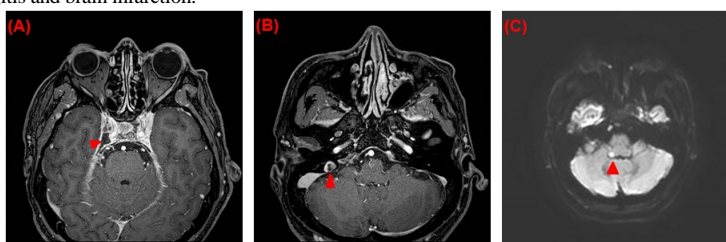


A report of rare and serious complications of atypical Lemierre's syndrome.

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Introduction: Lemierre's syndrome is a rare disease characterized by septic thrombophlebitis of the internal jugular vein with septicemia following an acute oropharyngeal infection. If the disease remains undiagnosed and untreated, it can result in severe complications and even death. We report a patient who developed rare complications of Lemierre's syndrome. **Case presentation:** A 48-year-old man was admitted with 2 weeks history of fever and right frontotemporal headache. He had poorly controlled diabetes. He was treated with periodontal infection for the previous 6 days without improvement. He had no meningeal irritation sign and CSF exam revealed no abnormality. His brain MRI and transesophageal echocardiography showed no definitive abnormality. Blood cultures grew *Streptococcus constellatus* in 2/3 bottles. Ceftriaxone was continued, however his headache worsened. Suddenly diplopia and right-sided ptosis developed at hospital day 5. His mental status deteriorated to confusion. The repeated CSF exam revealed pleocytosis (15,530/ml, neutrophil 91%) and orbit MRI revealed the thrombophlebitis of bilateral cavernous sinuses and right IJV and diffuse meningeal enhancement. The septic thrombophlebitis of cavernous sinuses and meningitis was diagnosed and atypical presentation of Lemierre's syndrome was considered. Meropenem, teicoplanin and low molecular weight heparin was administered. Two days later, the patient suddenly showed ataxia and nystagmus. Brain MRI demonstrated the multifocal acute infarctions including right lateral medulla. Repeated echocardiography showed no vegetation. After four weeks of antibiotic treatment, brain imaging showed recanalization of right IJV and cavernous sinus. He was discharged on warfarin. **Conclusion:** Because Lemierre's syndrome can present with nonspecific symptoms, high index of suspicion is very important. When the cavernous sinus is additionally involved, serious CNS complication can happen. Repeated CSF examination and brain imaging is crucial when the headache aggravates despite initial treatment. The delayed diagnosis can result in severe bacterial meningitis and brain infarction.



(A) Cavernous sinus thrombosis
(B) Internal jugular vein thrombosis
(C) Brain infarction

Kikuchi disease, presented with subacute necrotizing lymphadenitis of skin biopsy

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Kikuchi disease is benign condition of unknown cause, characterized by cervical lymphadenopathy and fever. Diagnosis is confirmed by Biopsy of Lymph node. Characteristic features include paracortical areas of necrosis, abundant karyorrhexis and mononuclear cells reaction. Other manifestations include Cutaneous manifestations, Arthritis, Aseptic meningitis, Fatigue, etc. It has been reported cutaneous manifestations present as macules, papules, plaques, nodules, etc. Skin biopsy showed vacuolar interface changes, necrotic keratinocytes, lymphohistiocytic infiltration, karyorrhexis, etc. There were some skin biopsy of lymphohistiocytic infiltration, but there were no skin biopsy of Necrotizing subacute lymphadenitis. We found a person with skin biopsy of lymphohistiocytic infiltration, most consistent with Necrotizing subacute lymphadenitis. A 58-year-old man who was diagnosed with HTN, Asthma presented to our department. He complained of Fever for 1month and pain from Lt. elbow to 4th/5th finger. He visited other hospital 3 weeks ago and had taken Chest CT, Abdomen CT, Neck CT, C-spine MRI, TTE. CT scan showed necrotic LN in celiac area, r/o Tb, Metastasis, LNE of Rt. Hilar, lower paratracheal and subcarinal area, r/o lymphoproliferative disease, Central disc herniation combined OPLL in C3-T2 and severe central spinal stenosis in C4-5, C5-6, C6-7. TTE showed No RWMA and No vegetation. CBC revealed no Leukocytosis. When he visited outclinic patient, he had papule on his chest and back. So we did Skin biopsy on his back. After a day he was admitted. We did PET-CT and the result was similar to the previous CT. We also did TBLB & EBUS. Biopsy result was histiocytes c apoptotic bodies, suggestive of subacute necrotizing lymphadenitis. We diagnosed him as Kikuchi disease and start Prednisolone 20mg(day), 10mg(Night). After that, Fever was subsided. But IGRA test was positive, so we needed to rule out Tb lymphadenitis. Laparoscopic biopsy, targeting Celiac LN was done and Biopsy result was also subacute necrotizing lymphadenitis. It's a rare case, but We should alert to skin lesion which can be the clue of diagnosing Kikuchi disease, so We recommend to do skin biopsy.

