CASE REPORT

Lemierre Syndrome: An Overlooked but Re-surfacing Disease

LAIBA ZAIN, UZMA SIDDIQUE, HIRA NAWAZ, AASMA KAYANI, MURTAZA ALI GOWA

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Correspondence to:

Dr. Hira Nawaz,
Senior Registrar,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

E-mail: hiranawaz@hotmail.com

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ABSTRACT

Lemierre's syndrome is a rare but severe head and neck condition that typically begins as an upper tract infection, and may progress to septic thrombophlebitis. The internal jugular vein is classically affected, although extension to the facial veins and respiratory tract can occur. It usually develops as a complication of a bacterial pharyngeal infection and can lead to serious systemic consequences, including bacteremia and septic embolization. Despite advances in antimicrobial therapy, recent literature suggests that the prognosis remains poor. Management relies primarily on prompt antibiotic therapy, as no disease-specific guidelines exist, and the role of therapeutic-dose anticoagulation remains controversial.

We report a pediatric case of Lemierre's syndrome in a 9-year-old boy who presented with fever and respiratory distress, with imaging demonstrating extensive venous thrombosis and septic embolic complications. This case underscores the importance of early recognition and timely intervention in reducing morbidity and improving outcomes in this potentially fatal condition.

Key Words: *Lemierre syndrome, Pediatrics, Infectious disease*

INTRODUCTION

This condition was first described by Courmont and Cade in 1900, by Schottmuller in 1918, and by Dr. Andre Lemierre (a French microbiologist) in 1936, reporting cases involving 20 patients with pharyngeal tonsillitis or septic thrombophlebitis of the tonsils and peritonsillar vessels secondary to sepsis.[1-3]

The most common bacteria causing the disease are Fusobacterium necrophorum (an active anaerobic gram-negative bacillus) and Fusobacterium nucleatum. These bacteria can cause infection with a variety of toxins, including endotoxins and exotoxins. Other studies have also reported bacteria such as Streptococcus, Bacteroidetes, Staphylococcus aureus, Klebsiella pneumoniae, etc.[4]

The most common target of infection is the ENT (pharyngitis or tonsillitis), but it can also occur in other diseases of the head and neck, such as otitis media and mastoiditis.[5] Local venous thrombophlebitis occurs, eventually leading to internal jugular vein thrombosis. If untreated, severe infection and sepsis with multiple end-organ failure, with the lungs being the most common metastatic disease, may occur. Clinical history, physical examination, microbial isolation, and radiographic evidence of thrombosis are used to identify patients with Lemierre syndrome. It is characterized by (i) a history of recent

oropharyngeal infection, (ii) clinical or radiological evidence of internal jugular vein thrombosis (IJVT), and (iii) isolation of bacteria. A fourth test is often used as imaging, usually with venography, doppler of the cervical arteries, and MRI (and MRV), in addition to a CT scan of the lungs. Prompt recognition and rapid initiation of antibiotics cannot be overemphasized.[6]

CASE REPORT

A 9-year-old boy presented with an 8-day history of fever and a 6-day history of cough, with recurrent episodes over the preceding 3 months. He also had right-sided earache with frequent purulent discharge and deviation of the right eye. He was initially hospitalized at a local facility, where he underwent ear surgery and received intravenous antibiotics; however, due to clinical deterioration, he was referred to our center.

On presentation, the patient was in respiratory distress. Chest examination and radiography were suggestive of pneumothorax, and a left-sided chest tube was inserted with high-flow oxygen, leading to clinical stabilization. Neurologically, he was conscious with a Glasgow Coma Scale score of 15/15. Ophthalmological examination revealed abnormalities in the right eye. He was a known case of cleft lip and palate, with cleft lip repair performed four years earlier. Laboratory investigations performed on admission and during hospitalization are summarized in **table 1.**

TABLE 1. Initial laboratory findings at the time of diagnosis

Hemoglobin	8.9 g/dl
Total leucocyte count	18.9
Platelets	112
Pt	12.5 sec
APTT	23.6 sec
CRP	200
Calcium	8.1
Magnesium	2.2
Phosphorus	4.6
Sodium (Na)	132
Potassium (K)	3.9
Chloride (Cl)	99
Bicarbonate	27
Serum creatinine	0.5
Blood culture	Streptococcus species
Pleural fluid culture	No growth after 5 days of incubation
Lymphocyte immunophenotyping	Low absolute count of CD3+ total lymphocytes. Both subset of T-lymphocytes (CD4+ and CD8+) are decreased. CD4+/CD8+ ratio is maintained.CD19+ Total B-lymphocytes are within normal limits. CD56+ Natural killer cells are decreased.

Given the suspicion of Lemierre disease, further imaging was performed. High-resolution CT of the temporal bone showed right-sided mastoiditis with thrombus involving the ipsilateral sigmoid sinus **(fig 1 & 2).** CT of the chest demonstrated bilateral consolidation with interstitial septal thickening and multiple thick-walled air-filled cavitations, consistent with septic pulmonary emboli **(fig 3).** MRI brain with MR venography confirmed right-sided otomastoiditis with thrombosis of the right sigmoid and transverse sinuses extending into the right internal jugular vein, with mild mucoperiosteal thickening in the bilateral ethmoid air cells **(fig 4).**

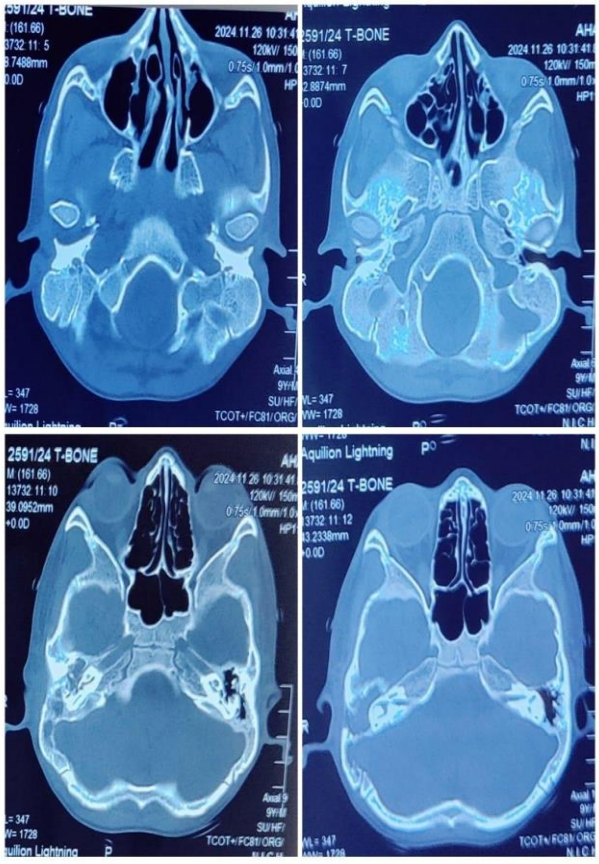

Fig 1: Axial view of hrct temporal bone: showing

right sided mastoiditis and thrombus involving ipsilateral sigmoid sinus

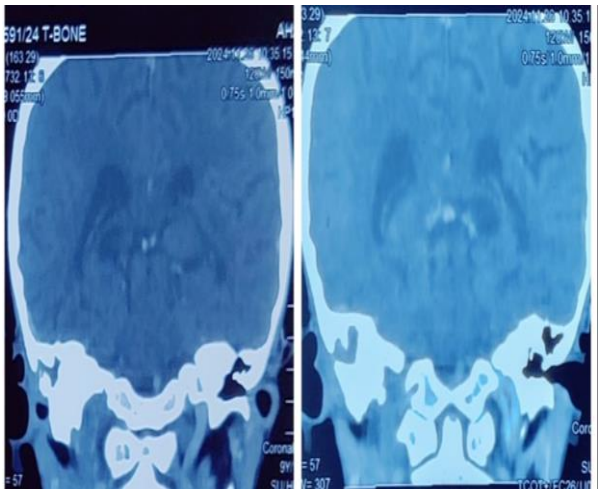

Fig 2: Coronal view of temporal bone

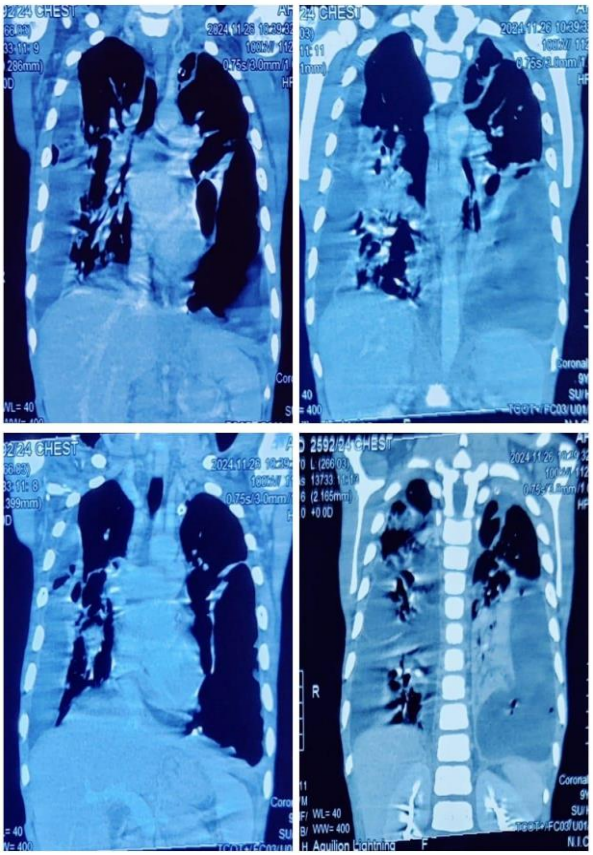

Fig 3: CT chest axial and coronal images showing bilateral consolidation with interstitial septal thickening and multiple thick walled air filled cavitations

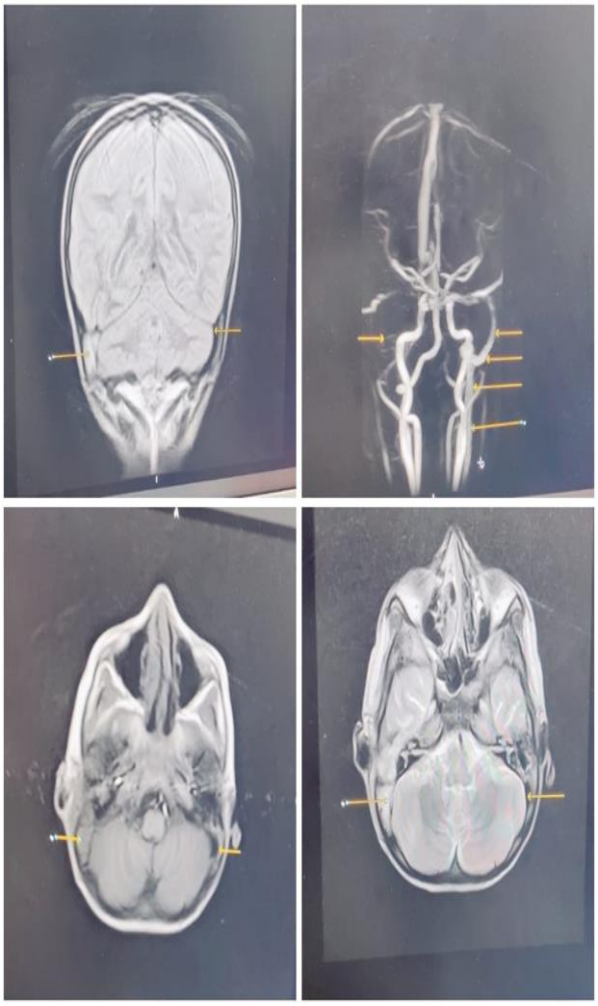

Fig 4: MRI brain with mrv showing right sided oto-mastoiditis with right sigmoid and right transverse sinus thrombosis extending to right internal jugular vein, mild mucoperiosteal thickening in bilateral ethmoid ear cells.

DISCUSSION

Lemierre syndrome (LS) is a rare complication of ENT infection resulting from IJV-infectious thrombophlebitis.[7] Andre' Lemierre stated that sepsis and distal septic emboli may arise from the nasopharynx, oral cavity, palate, otitis media and urethra, or from mastoiditis, purulent endometritis and appendicitis.[8] Although LS is usually seen in adolescence or young adults, it can also be seen in childhood, as seen in our case. Due to its rarity and lack of obvious symptoms, LS is difficult to

diagnose. In addition, this rare event is associated with serious complications from ENT infection, as the infection can develop rapidly and sometimes lead to sepsis, pneumonia, or other life-threatening problems.[7] Although IJV thrombophlebitis is a key symptom of LS, these symptoms are not required for diagnosis of the disease. Evidence of IJV thrombophlebitis or evidence of metastatic disease at the site, including isolation of F. necrophorum from blood or other sterile sources.[9]

LS can rapidly progress to disseminated disease because pathogenic organisms can bypass mucosal defenses due to local inflammation, injury, or tissue damage. Once bacteria enter the head and neck vasculature, they can form a thrombotic site and spread to distant organs via septic emboli. The lung is the most common site for septic emboli, but metastatic diseases such as septic arthritis, osteomyelitis, meningitis, pericarditis, and liver abscesses can also occur.[9]

A 2020 literature review found that movement disorders (characterized by diplopia and paralysis of the III, IV, and VI cranial nerves) were the most common symptoms and that ophthalmic problems occurred in 88.89% (24 cases) of LS cases: According to the method, nerve palsy is seen in 50% of LS patients with no disease. body alignment (12 patients) and ophthalmic problems are seen in 44.44% of LS patients.[9]

LS treatment is threefold, based on intensive antibiotics, immunotherapy and surgery.[9,10] Studies have shown that all F. necrophorum strains are sensitive to metronidazole, ticarcillin-clavulanic acid, cefoxitin, amoxicillin/clavulanic acid and imipenem, while resistance to erythromycin occurs in 15% to 22% of patients.[9] Metronidazole appears to be the best antibiotic for the treatment of human necrotobacteriosis due to its good oral bioavailability, good protection against Fusobacterium spp. and strong tissue and central nervous system activity.[9] When metronidazole was given together with carbapenems or piperacillin/tazobactam, the success rate was up to 98% over a 4-week treatment period. If patients improve after two to three weeks of intravenous antibiotic therapy, they can switch to oral antibiotics, usually metronidazole, for another four weeks.[9]

Anticoagulant therapy is often recommended because it increases the rate of thrombus destruction and increases the penetration of antibiotics into septic emboli.[9] A 2002 meta-analysis of the effect of anticoagulation on sinus thrombosis found no significant reduction in mortality.[9] A prospective study published in 2004 demonstrated that heparin therapy achieved an 80% success rate in patients with sinus thrombosis, with 79% of patients recovering, 5% experiencing serious complications, and 8% mortality.[9] These findings have contributed to heparin becoming a standard component of care in many neurological thrombotic disorders. Based on such evidence, most neurologists favor anticoagulation alongside antibiotic therapy in appropriate cases, particularly when complications arise that may otherwise require surgical intervention.

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Authors' affiliation

Dr. Laiba Zain,
Resident,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

Dr. Uzma Siddique,
Fellow,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

Dr. Hira Nawaz,
Senior Registrar,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

Dr. Aasma Kayani,
Fellow,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

Dr. Murtaza Ali Gowa
Associate Professor & Section Head,
Department of Pediatric Intensive Care Unit, National Institute of Child Health, Karachi, Pakistan

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Authors' contribution

LZ: Proposed topic, basic study design, methodology and manuscript writing
US: Data collection, statistical analysis and interpretation of results etc.
HN: Literature review & referencing and quality insurer
AK: Clinical care & data collection
MAG: Critical revision

All the authors have approved the final manuscript draft and accept the responsibility of research integrity.