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Case Report

A Lemierre's like syndrome in a 50 days old infant with Enterobacter cloaca bacteremia: A case report☆,☆☆

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ABSTRACT

Lemierre syndrome describes septic thrombophlebitis of the internal jugular vein (IJV) and metastatic spread of the infection following a recent oropharyngeal infection in a setting of bacteremia caused by *Fusobacterium necrophorum*. Lemierre-like syndrome describes similar clinical scenario with no preceding oropharyngeal infection and/or in the setting of non-Fusobacterium cause. We report a case of Lemierre-like syndrome in a setting of Enterobacter cloaca bacteremia without known preceding oropharyngeal infection. History and physical exam revealed an irritable infant with cough, tachypnea, low grade fever, bilateral lung crepitations and features of infantile seborrheic dermatitis on the scalp. Imaging revealed thrombosis of right internal jugular vein and superior vena cava, bilateral pulmonary cavitary lesions and collections consistent with septic pulmonary emboli. Multiple rim enhancing hypo-dense liver lesions and chest wall collections consistent with abscesses were also seen. He was managed with parental antibiotics, drainage of the chest wall abscesses and discharged with clinical and radiologic improvement.

Introduction

Lemierre's syndrome is characterized by jugular vein thrombosis (JVT) associated with suppurative anaerobic infection of the upper aero-digestive tract, often complicated by septic pulmonary embolism (PE). The usual etiologic agent is Fusobacterium necrophorum; a pleiomorphic Gram-negative bacillus identified from anaerobic culture of blood or purulent fluid [1,2]. The constellation of findings is called Lemierre-like

Abbreviations: LS, Lemierre syndrome; LLS, Lemierre-like syndrome; IJV, Internal jugular vein; CT, Computed tomography; ESR, Erythrocyte sedimentation rate; SVC, superior vena cava.

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syndrome (LLS) when Fusobacterium is not the agent responsible or/and the source of infection is not oropharyngeal [3,4]. It has historically been regarded as a disease of healthy adolescents or young adults and occurs at an overall rate of 1 to 10 cases per million person-years with an estimated fatality rate of 4% to 9% [5]. The incidence and mortality rate has seen a dramatic reduction in the postantibiotic era. However, In recent years there is increase in published cases, including in children and still carry a mortality rate of 2%-8% [6–8]. A systematic review in 2021 that included 712 patients with the condition reported that the median age was 21 (inter-quartile range = 17-33) [9]. We describe a case of Lemierre like syndrome in a 50 days old infant. To the best of our knowledge, this case is among the youngest described in the literatures.

Case presentation

A 50 days old male term infant presented with fast breathing, cough and high grade intermittent fever of 10 days duration. He also had scalp swelling over the occipital area which occurred 2 days prior to onset of other symptoms. He was referred to our institution from a nearby primary hospital. Initially, he visited this primary hospital for small posterior scalp lesion that was oozing pus at the time. He was admitted there for 7 days being treated empirically with IV ampicillin and gentamicin for the diagnosis of late onset neonatal sepsis. Central venous catheter placement was not attempted. No prior history of illness or hospital visit. At the time of presentation to our institution, he was irritable and exhibiting signs of respiratory distress. He was tachypnic (respiratory rate = 80/minute), febrile (temperature = 37.9°C), tachycardic (pulse rate = 150 beats/minute) and oxygen saturation (SpO2) level was at 76% with a room air oxygen. Chest Examination revealed sub-costal and inter-costal retractions, nasal flaring and also bilateral lower lung field coarse crepitations. There was a 5cm x 3cm superficial scalp raised lesion over the occipital area having crusted yellowish surface. There were multiple mobile small flocculent soft-tissue swellings involving the anterior and posterior chest wall. No abdominal distention.

The laboratory examinations done on the same day revealed elevated white blood cell count 27,900 cells/μL, hemoglobin 9.1 mg/dL and erythrocyte sedimentation rate (ESR) 60 mm/hour. Peripheral Blood smear revealed normocytic, normochromic RBC and no blast cells seen. At the time, neck ultrasound showed no evidence of retropharyngeal collection or venous thrombosis. Microscopic analysis of the aspirated chest wall collection suggested chest wall abscess having sheets of inflammatory cells, predominantly neutrophils on necrotic background. On the chest radiograph there were bilateral multiple randomly distributed nodular airspace opacities involving all lung zones (Fig. 1A). With initial diagnosis of Bronchopneumonia and infantile seborrheic dermatitis, the patient was put on supplemental intranasal oxygen and empirical treatment was initiated with vancomycin 10 mg/kg/dose q6hr and ceftazidime 30 mg/kg IV q8hr, awaiting further investigation.

Computed tomography (CT) scan of the chest and abdomen done 6 days after admission showed multiple bilateral predominantly peripheral well-defined rim enhancing, centrally fluid attenuating Nodules involving all lung segments with feeding vessel sign (Figs. 1B-E). Some of the lesions demonstrated central cavitation (Figs. 1A and 2B). Separate rim enhancing lesions involving the anterior and posterior chest wall soft tissue is seen. There was also few thin walled air filled cystic sub-pleural lesions in the right lung upper lobe anterior segment. Free air was seen in the left pleural cavity predominantly occupying the nondependent regions (Figs. 1B and 2). But no pleura space loculated fluid seen. There was also nonenhancing filling defect involving the distal segment of the right internal Jugular and Sub-Clavian veins and also the superior vena cava (SVC). Multiple round, centrally hypodense, peripherally enhancing lesions were seen scattered in the right liver lobe segments (Figs. 3 and 4). The diagnosis of septic pulmonary embolism with chest wall abscesses, left side pneumothorax, right side proximal internal jugular and subclavian veins and SVC thrombosis was made. Further neuro-imaging studies was not done considering the patient did not show clear signs of neurologic impairment.

Culture from the scalp lesion do not show growth. However, Enterococcus cloaca was isolated from the blood culture sample. The organism was sensitive to meropenem (see Table 1) and the antibiotic choice was revised to meropenem 20 mg/kg IV q8hr. Subsequently the patient showed significant clinical improvement at 16th day of admission with reduction in respiratory rate, pulse rate and temperature and maintained normal oxygen saturation without supplemental oxygen. Chest radiograph repeated at 21th day of admission showed significant reduction in air-space opacities bilaterally compared with the initial radiograph (Fig. 5). Finally, after 25 days of hospital stay the infant was discharged improved.

Table 1 – Drug susceptibility test and zone of inhibition for the isolated Enterobacter cloaca species.

Drug	Zone size (mm)	Interpretation (S, I, R)	Drug	Zone size (mm)	Interpretation (S, I, R)
Ampicillin	6	R	Cefotaxime	6	R
Cefazolin	6	R	Ciprofloxacin	16	R
Gentamicin	6	R	Imipenem	26	S
Amoxicillin/Clavulanate	19	S	Amikacin	18	S
Piperacillin-Tazobactam	22	S	Meropenem	27	S
Trimethoprim/Sulfa	6	R	Cefepime	6	R
Chloramphenicol	21	S	Ceftriaxone	6	R

I, Indeterminate; S, sensitive; R, resistant.

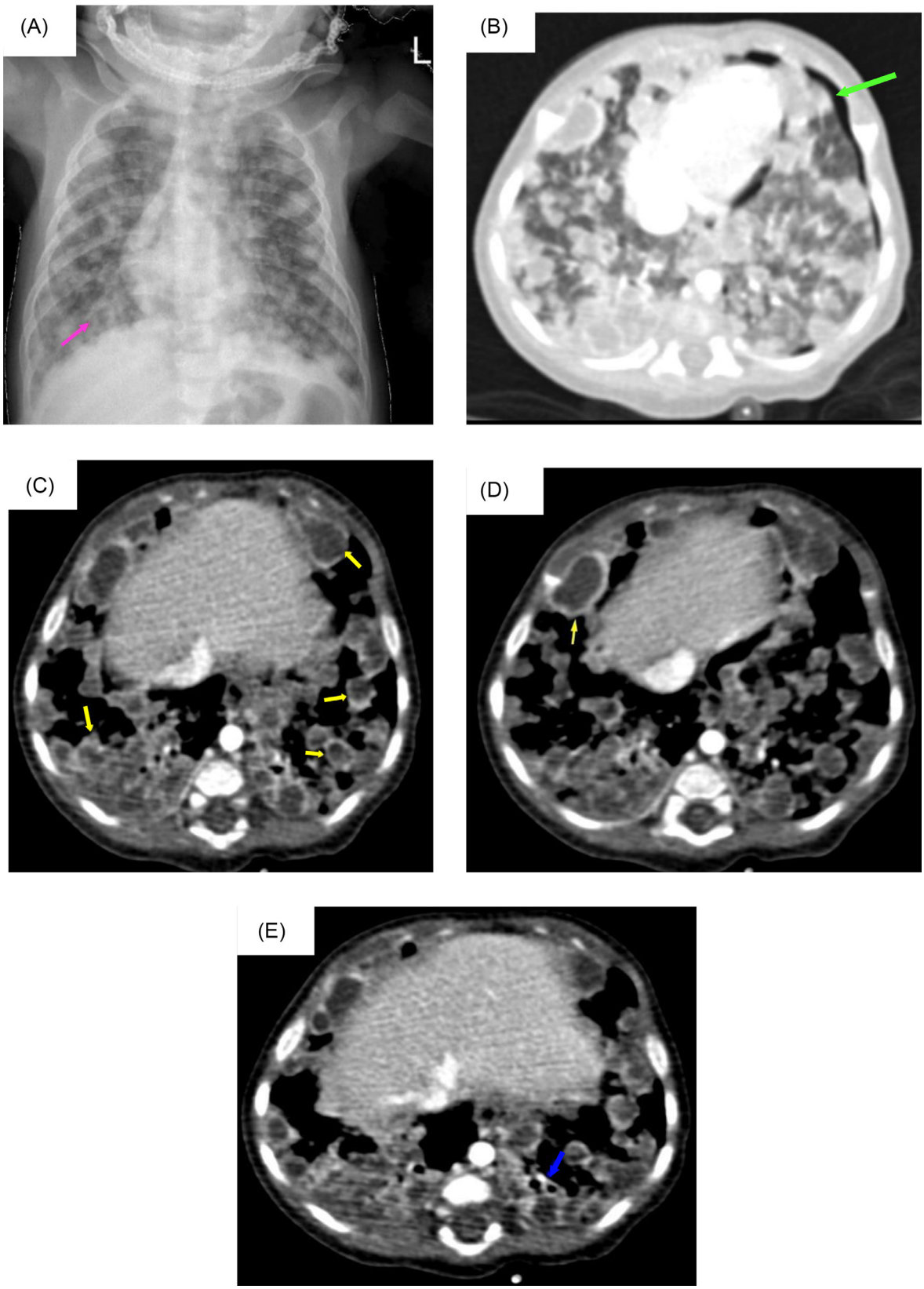

Fig. 1 – (A) Chest radiograph taken at admission shows multi-focal bilateral randomly distributed nodular air space opacities, some having central cavitation (pink arrow). (B) Chest CT, axial lung window showing multiple nodular opacities and left side pneumothorax (green arrow). (C-E) Chest CT, Axial soft tissue window images showing multiple, predominantly peripheral ring enhancing collections (yellow arrow) and feeding vessel sign (blue arrow).

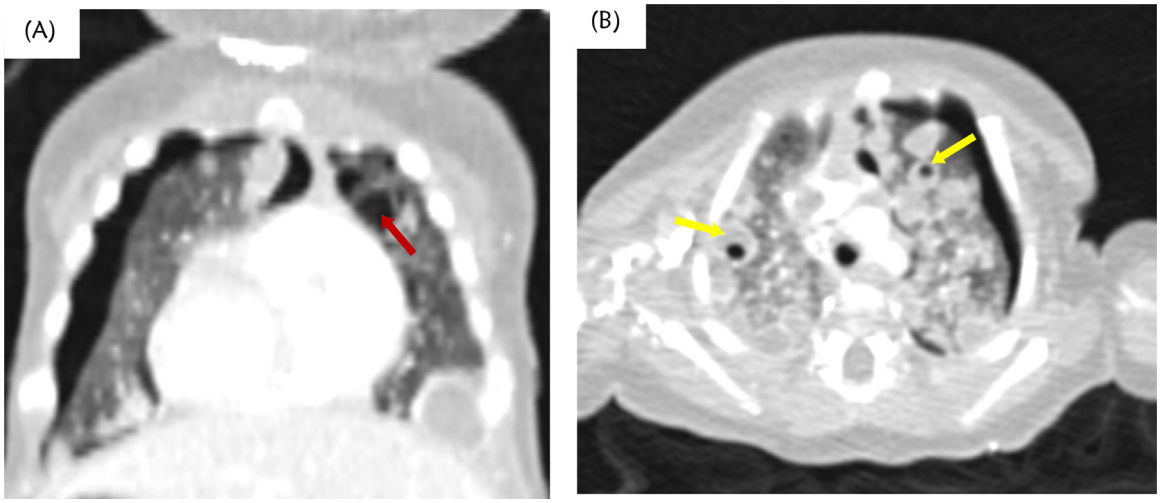

Fig. 2 – (A) Chest Ct, coronal, lung window showing right side pneumothorax and left upper lobe apical segment pulmonary cysts (Brow narrow). (B) Chest CT, Axial Lung window showing bilateral cavitating nodules (Yellow arrows).

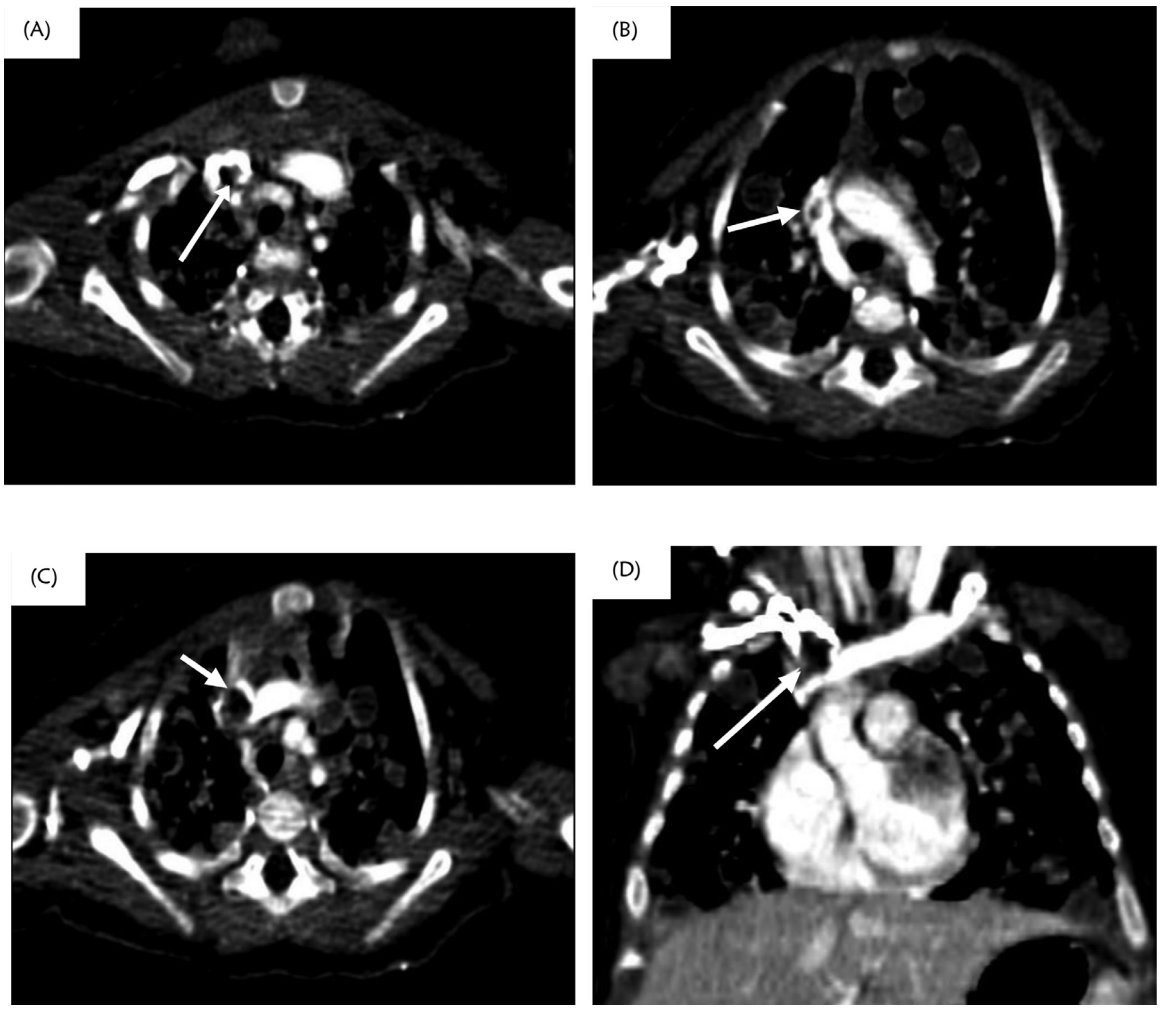

Fig. 3 – Chest Ct, axial (A-C) and coronal (D) postcontrast images showing central filling defect with enhancing periphery indicating thrombophlebitis in the proximal right internal jugular vein (IJV) (White arrow in [A]) and the distal superior vena cave (White arrow in [B-D]).

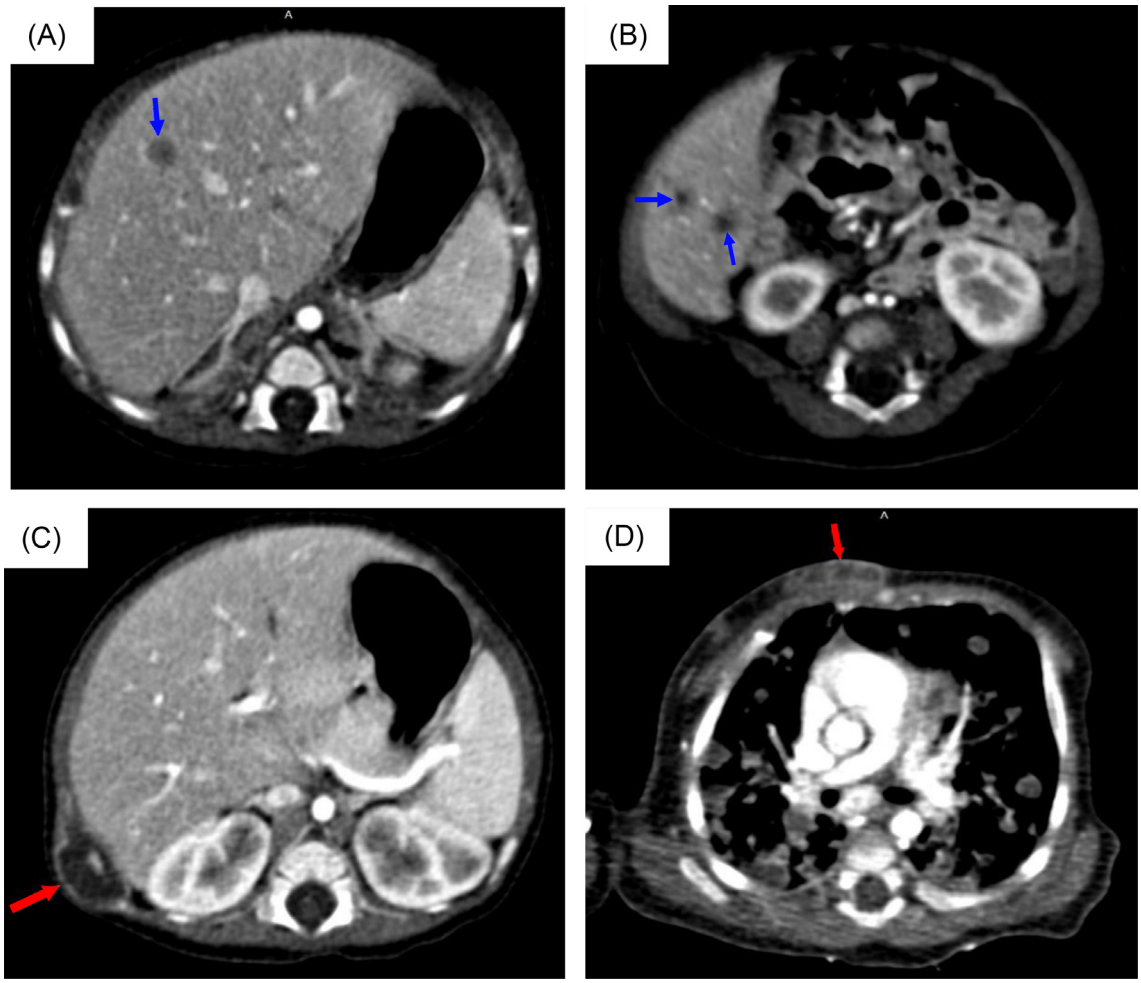

Fig. 4 – (A-D) Axial postcontrast CT of the chest and upper abdomen showing hypo-dense rim enhancing collections in the anterior and lateral chest wall (red arrows) and multiple hypo-dense peripherally enhancing lesions in the right liver lobe (blue arrows).

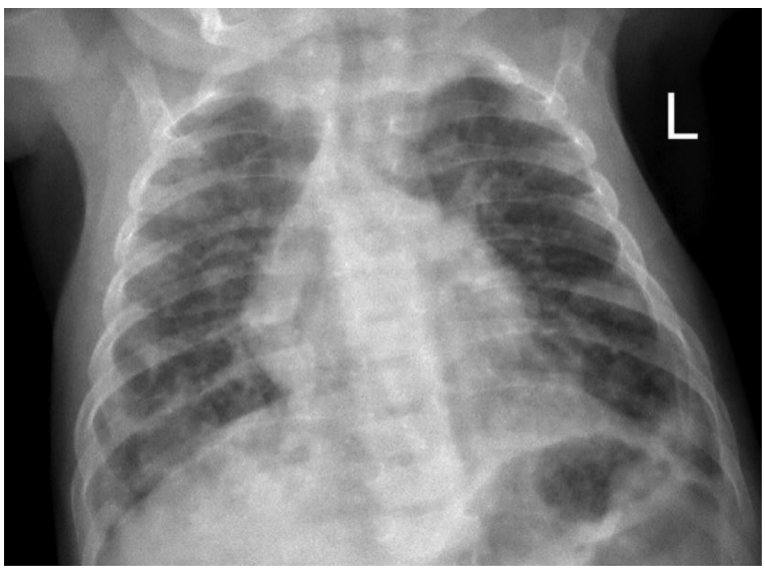

Fig. 5 – Chest radiograph repeated after 21 days shows remarkable reduction in the opacities bilaterally compared to the initial radiograph (see Fig. 1A).

Discussion

Lemierre syndrome (LS), first described in 1936 by Dr Andre Lemierre (a French microbiologist), describes septic thrombophlebitis of the internal jugular vein (IJV) and metastatic spread of the infection following a recent oropharyngeal infection in a setting of bacteremia caused by anaerobic organisms, mainly *Fusobacterium necrophorum*. A similar clinical syndrome with no preceding oropharyngeal infection and/or in the setting of non-Fusobacterium origin is described by many as Lemierre-like syndrome (LLS) [3]. In the absence of internal jugular vein thrombophlebitis, the presence of one or more metastatic lesions is considered as a thrombophlebitis marker [10]. Etiologic agents other than Fusobacterium associated with LS or LLS include Streptococcus, Staphylococcus, Enterococcus, Bacteroides, and Lactobacilli. Those microorganisms may be cultured alone or in combination with *F. necrophorum* [11]. Enterococcus cloaca is among pathogens commonly incriminated in nosocomial infections in neonatal intensive care units (ICU) [12]. Primary source of infection in LLS, with nonoropharyngeal source include Chest infection, Middle ear/mastoid, Orbit, gastrointestinal and skin [13]. In our case, the initial proposed site for the source of infection was the scalp lesion, for which he was admitted to nearby primary hospital. However, even though sample was taken after a course of empirical antibiotics, absence of growth on the culture from the scalp lesion did not support this claim. But he was taking IV antibiotic at this primary hospital for 7 days and this can be possible source of nosocomial infection for our patient.

After being considered "the forgotten disease" for so long in post antibiotic era, there has been an increase in reporting of pediatrics LS and LLS over the past 2 decades. Weather this reflects a true increase in the incidence of the syndrome or just a publishing trend remains to be seen [14,15]. To the best of our investigation, So far the youngest reported cases of LS and LLS are a 5-week-old male with peritonsillar/parapharyngeal preceding LS and a 2-month-old boy with preceding otomastoiditis. This makes our case among the youngest patients with LLS at just 50 days [16].

Thrombophlebitis initially occurs in the draining veins near the source of infection and spreads to the internal jugular vein, where platelet aggregation among other factors contributes to the formation of infected thrombus. Septic emboli then seed to the lung capillaries and subsequently to distant organs to form metastatic abscesses [17]. The most frequently involved site of septic metastases are the lungs, followed by the joints (knee, hip, sternoclavicular joint, shoulder, and elbow). Other sites involved in septic metastasis and abscess formation are the muscles and soft tissues, liver, spleen, kidneys, and central nervous system [13]. Septic emboli in the lung can result in pneumothorax, which is a rare but life threatening complication of LLE [18]. This makes it one of the key findings but rare finding seen in our case.

The mechanisms of thrombus formation in LS and LLS are not completely understood. Suggested mechanisms include direct cell-to-cell contact of the organism with platelets resulting in platelet aggregation and thrombosis. Another proposed mechanism is presence of transient acute inflammatory pro-thrombotic state characterized by antiphospholipid antibodies and elevated factor VIII levels. A study of thromboembolic outcomes of LS and LLS in children concluded the presence of transient acute inflammatory prothrombotic state rather than intrinsic hypercoagulability contributing to the development of IJV thrombosis. The absence of clear imaging evidence of phlebitis in our case favors the explanation in the second mechanism [6].

Antibiotic therapy has been an integral part of the management. Choice of antimicrobial agents should be individualized according to antibiotic sensitivity. The initial empiric antibiotic regimen should cover Fusobacterium Necrophorum and other potential pathogens involved. In the presence of a lung abscess, the use of piperacillin-tazobactam, meropenem, or second-generation cephalosporin plus metronidazole as empiric regimen has been suggested. The duration of treatment usually ranges from 10 days to 8 weeks [2,19]. Given the rarity of the condition, absence of adequate multi-centric randomized clinical trials, and the risk associated, the role of anticoagulants in treating Lemierre's syndrome remains controversial and the decision should be based on the individualized scenario [19].

Classic Lemierre syndrome includes 4 findings: primary oropharyngeal infection, bacterial sepsis with at least 1 positive blood culture, clinical or radiographic demonstration of thrombosis in a vein of the head or neck, and at least 1 distant site of infection. Our patient's presentation is consistent with Lemierre-like syndrome (LLS) where enterococus cloaca is the isolated microorganisms from blood culture sample. Enterococus cloacae is one of the most frequently encountered human pathogens among the genus Enterobacter. It is a Gram-negative bacterium that resides as a normal flora in the human gastrointestinal tract. It is also among increasingly important nosocomial pathogens. Although majority of infections with Enterobacter spp. are nosocomial, community-acquired infections with this organism do occur [20]. The antibiotic sensitivity has been crucial for the success in managing this case because the organism, enterococus cloaca is known to have resistance to multiple antimicrobials and is commonly incriminated pathogen in health care associated outbreaks.

Patient consent

After a thorough explanation of the case report and its publication process, written informed consent for the inclusion of their child's medical information was obtained from the patient's parents. This ensures their understanding and authorization for the anonymized presentation of this case for educational purposes.

REFERENCES

[1] Lemierre A. On certain septicæmias due to anaerobic organisms. Lancet 1936;227:701–3.
[2] Kuhajda I, Zarogoulidis K, Tsirgogianni K, Tsavlis D, Kioumis I, Kosmidis C, et al. Lung abscess-etiology,

diagnostic and treatment options. Ann Transl Med 2015;3(13):183. doi:10.3978/j.issn.2305-5839.2015.07.08.
[3] Van Hoecke F, Lamont B, Van Leemput A, Vervaeke S. A Lemierre-like syndrome caused by Staphylococcus aureus: an emerging disease. Infect Dis (Lond) 2020;52(3):143–51. doi:10.1080/23744235.2019.1691255.
[4] Newman N, Bantikassegn A, West TG, Peacock JE Jr. An unusual etiology of lemierre-like syndrome: preseptal cellulitis due to methicillin-resistant *Staphylococcus aureus*. Open Forum Infect Dis 2022;9(5):ofac143. doi:10.1093/ofid/ofac143.
[5] Sacco C, Zane F, Granziera S, Holm K, Creemers-Schild D, Hotz MA, et al. Lemierre syndrome: clinical update and protocol for a systematic review and individual patient data meta-analysis. Hamostaseologie 2019;39(1):76–86. doi:10.1055/s-0038-1654720.
[6] Goldenberg NA, Knapp-Clevenger R, Hays T, MJ Manco-Johnson. Lemierre's and Lemierre's-like syndromes in children: survival and thromboembolic outcomes. Pediatrics 2005;116(4):e543–8. doi:10.1542/peds.2005-0433.
[7] Karkos PD, Asrani S, Karkos CD, Leong SC, Theochari EG, Alexopoulou TD, et al. Lemierre's syndrome: a systematic review. Laryngoscope 2009;119(8):1552–9. doi:10.1002/lary.20542.
[8] Johannesen KM, Bodtger U. Lemierre's syndrome: current perspectives on diagnosis and management. Infect Drug Resist. 2016 Sep 14;9:221-227. doi: 10.2147/IDR.S95050.
[9] Valerio L, Zane F, Sacco C, Granziera S, Nicoletti T, Russo M, et al. Patients with Lemierre syndrome have a high risk of new thromboembolic complications, clinical sequelae and death: an analysis of 712 cases. J Intern Med 2021;289(3):325–39. doi:10.1111/joim.13114.
[10] García-Salido A, Unzueta-Roch JL, García-Teresa MÁ, Sirvent-Cerdá S, Martínez de Azagra-Garde A, Casado-Flores J. Pediatric disseminated lemierre syndrome in 2 infants: not too young for an ancient disease. Pediatr Emerg Care Jul 2017;33(7):490–3. doi:10.1097/PEC.0000000000000570.
[11] Chirinos JA, Lichstein DM, Garcia J, Tamariz LJ. The evolution of Lemierre syndrome: report of 2 cases and review of the literature. Medicine (Baltimore) 2002;81:458–65.
[12] Osman AH, Darkwah S, Kotey FCN, Odoom A, Hotor P, Dayie NTKD, et al. Reservoirs of nosocomial pathogens in intensive care units: a systematic review. Environ Health Insights. 2024;18:11786302241243239. doi: 10.1177/11786302241243239.
[13] Karkos PD, Asrani S, Karkos CD, Leong SC, Theochari EG, Alexopoulou TD, et al. Lemierre's syndrome: a systematic review. Laryngoscope 2009;119(8):1552–9. doi:10.1002/lary.20542.
[14] Lavalle S, Masiello E, Cocuzza S, Pavone P, Di Nora A, Calvo-Henriquez C, et al. Pediatric Lemierre's syndrome: a comprehensive literature review. Pediatr Rep. 2024;16(1):201-213. doi: 10.3390/pediatric16010018.
[15] Weesner CL, Cisek JE. Lemierre syndrome: the forgotten disease. Ann Emerg Med 1993;22(2):256–8. doi:10.1016/s0196-0644(05)80216-1.
[16] Raggio BS, Grant MC, Rodriguez K, Cripe PJ. Neonatal Lemierre syndrome: youngest reported case and literature review. Clin Pediatr (Phila) 2018;57(3):294–9. doi:10.1177/0009922817721156.
[17] Rae J, Misselbrook K. Lemierre's Syndrome: a rare cause of disseminated sepsis requiring multi-organ support. J Intensive Care Soc 2017;18(4):329–33. doi:10.1177/1751143717698978.
[18] Okabe M, Kasai K, Yokoo T. Pneumothorax secondary to septic pulmonary emboli in a long-term hemodialysis patient with psoas abscess. Intern Med 2017;56(23):3243–7. doi:10.2169/internalmedicine.9050-17.
[19] Gore MR. Lemierre syndrome: a meta-analysis. Int Arch Otorhinolaryngol 2020;24(3):e379–85. doi:10.1055/s-0039-3402433.
[20] Sanders WE Jr, Sanders CC. Enterobacter spp.: pathogens poised to flourish at the turn of the century. Clin Microbiol Rev 1997;10:220–41. doi:10.1128/CMR.10.2.220.