IMAGES IN THORAX

Unilateral proptosis in Lemierre's syndrome variant

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A 47-year-old man, with a 10-year history of alcohol consumption, presented with a left cheek swelling for 14 days following self-extraction of two teeth. On the day of admission, he developed a low-grade fever, delirium and left eye proptosis (figure 1A). Laboratory analysis showed a white cell count of 21.6×10^9 cells/L with 79.8% neutrophils. C-reactive protein was 26.3 mg/dL. Blood chemistries were within normal limits except creatinine 2.2 mg/dL and sodium 128 mmol/L. CT imaging of the brain revealed left superior and inferior ophthalmic vein engorgement (figure 1B), suspected cavernous sinus thrombosis (CST) related and a tissue swelling over left pterygoid muscles, consistent with an abscess (figure 1C, arrow). Chest X-ray revealed mass-like lesions over the left-upper and right-lower lung fields (figure 1D).

Eight days later, blood cultures grew *Fusobacterium necrophorum*. At this time, we made a diagnosis of a variant of Lemierre's syndrome based on septicaemia with *F. necrophorum* complicated with unilateral proptosis and septic pulmonary embolism in the absence of internal jugular vein (IJV) thrombosis. The patient had a response to therapy with metronidazole and ceftriaxone and was discharged on the 26th day after admission. We reviewed the patient after discharge with 4 weeks of oral clindamycin and noted complete recovery in his proptosis, acute renal failure and repeated chest radiographs.

In 1936, André Lemierre was the first person who described the syndrome with the following key components of (i) an acute oropharyngeal infection as initial presentation, (ii) progression to septic thrombophlebitis of the IJV and (iii) leading to septicaemia and metastatic disease.[1] The main causative organism is *F. necrophorum*, which is a gram-negative anaerobic bacillus and forms part of normal oropharynx microflora. However, these presentations are typical but not universal. Alternative primary sources of infection are sinusitis, otitis media and odontogenic infection. IJV thrombophlebitis is not always presented. Therefore, Kristensen and Prag define 'isolation of *F. necrophorum* with head and neck focus and clinical evidence of dissemination' as 'variants of Lemierre's syndrome'.[2] In this case, self-extraction of two teeth related mucosal damage and alcoholism-induced immune compromise may be the aetiologies of invasive *F. necrophorum* infections.

F. necrophorum septic emboli could cause widespread haematological damage to the central nervous system, joints, kidneys and liver. However, pulmonary involvement is most common. Proptosis is a rare central nervous system complication of Lemierre's syndrome, which is due to CST. CST can be aseptic but is usually septic. Septic CST is most commonly caused by *Staphylococcus aureus* or *Streptococcus species* related sinusitis. However, primary odontogenic infection with *F. necrophorum* could be a rare aetiology, as shown in our case.[3] The retrograde spread of septic metastases from the postanginal area via the sinovenous system is presumably the possible route.

In the era of widespread use of antibiotics, the incidence of Lemierre's syndrome has fallen to a degree of 'forgotten disease'. However, once *F. necrophorum* haematogenously spreads to multiple organs, Lemierre's syndrome is a potentially fatal disease. The case highlights

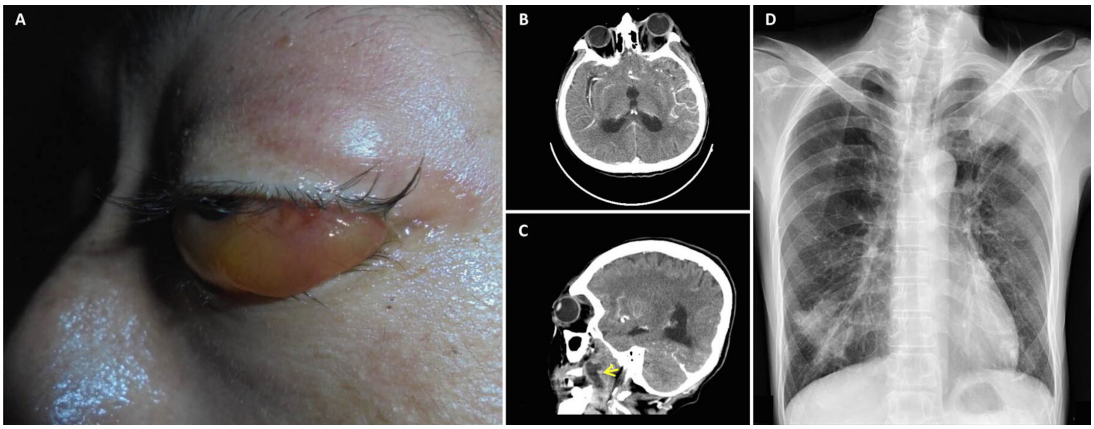

Figure 1 A 47-year-old man developed a variant of Lemierre's syndrome. (A) Left eye proptosis. (B) CT imaging of the brain revealed left superior and inferior ophthalmic vein engorgement, suspected cavernous sinus thrombosis related, (C) an abscess over left pterygoid muscles. (D) Chest X-ray revealed mass-like lesions over left-upper and right-lower lung fields.

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that a variant of Lemierre's syndrome should be suspected in a person who develops an oropharyngeal infection and then exhibits symptoms and signs in the week of pulmonary or central nervous system involvements. Chest radiographs, blood cultures and contrast-enhanced CT scanning should be ordered to provide a diagnosis.

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REFERENCES

1 Lemierre A. On certain SEPTICÆMIAS due to anaerobic organisms. *Lancet* 1936;227:701–3.
2 Hagelskjaer Kristensen L, Prag J. Lemierre's syndrome and other disseminated Fusobacterium necrophorum infections in Denmark: a prospective epidemiological and clinical survey. *Eur J Clin Microbiol Infect Dis* 2008;27:779–89.
3 Branson SV, McClintic E, Yeatts RP. Septic cavernous sinus thrombosis associated with orbital cellulitis: a report of 6 cases and review of literature. *Ophthalmic Plast Reconstr Surg* 2019;35:272–80.