From the dentist to the neurologist: a case of incomplete Lemierre syndrome due to odontogenic infection

P. Cerrone[1], P. Sucapane[1], S. Mearelli[1], R. Totaro[1], M. Baldassarre[1], S. Sacco[2], C. Marini[3]

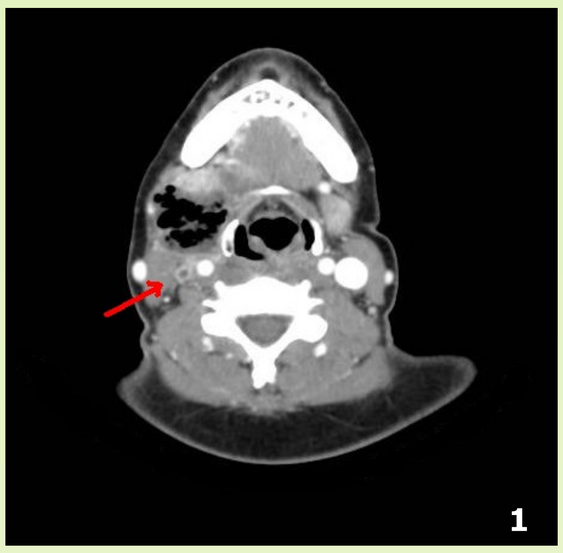
1 Neurology Unit, Hospital San Salvatore, L'Aquila
2 Neurology Unit, Hospital San Salvatore, Department of Biotechnological and Applied Clinical Sciences, University of L'Aquila
3 Neurology Unit, Hospital San Salvatore, Department of Life, Health and Enviromental Sciences, University of L'Aquila

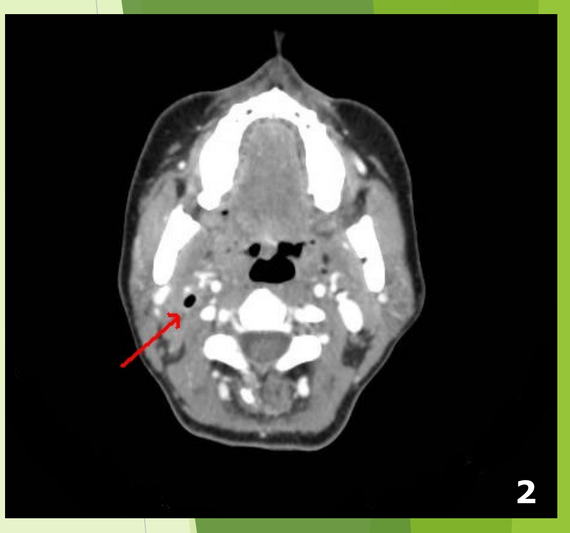
BACKGROUND

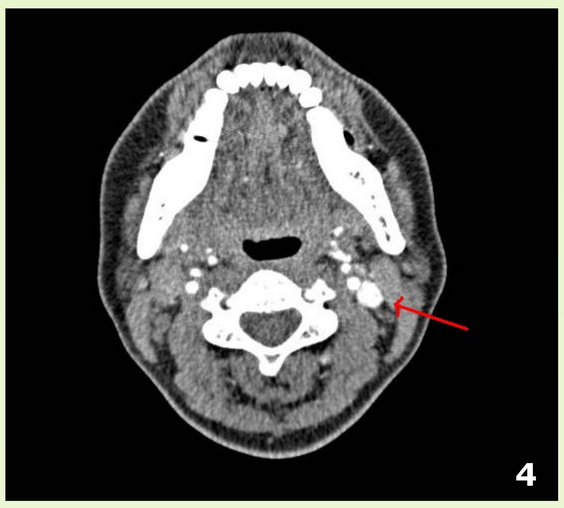
Lemierre syndrome (LS) is characterized by a history of recent oropharyngeal infection, clinical or radiological evidence of internal jugular vein thrombosis and evidence of metastatic lesions in lungs and/or another remote site. It is a rare but serious disease, often caused by a septic focus lying in the head and neck region. [1] Pharyngitis and tonsillitis are responsible for over 85% of LS cases, while mastoiditis and odontogenic infections are far less common, accounting for 3% and 2% of LS cases, respectively, as the site of primary infection. [2]
This syndrome was first described in 1936 by Lemierre. The introduction of antibiotics in the clinical practice caused a substantial decrease of the incidence and mortality of LS, which was termed "the forgotten disease."[1]
We describe a case of incomplete Lemierre syndrome due to an odontogenic infection.

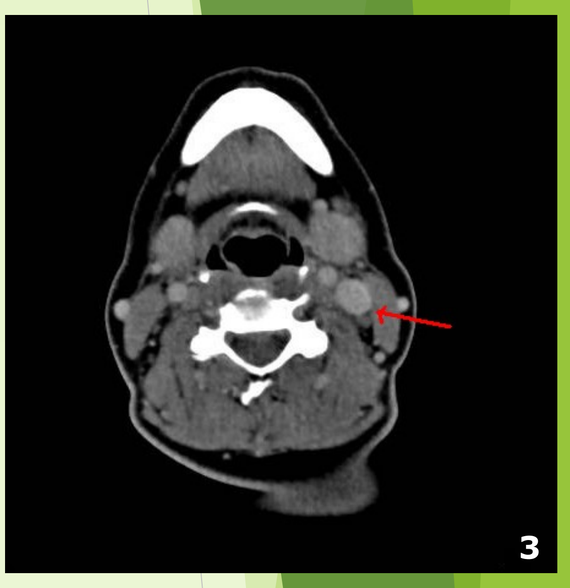
CASE REPORT

A 38-year-old woman presented to the local hospital complaining fever, local pain, headache and odynophagia. The patient underwent lower right third molar avulsion six months later, but dental removal was incomplete and resulted in the avulsion of crown with permanency of dental roots. Fifteen days before the admission the patient began to present local pain and inflammation with pyorrhea and a dentist gave her oral cephalexin. After a cycle of cephalexin the patient started oral ciprofloxacin 500 mg bid. At admission physical examination showed a submandibular swelling, spreading to the right side of the neck, pyorrhea from right retromolar trigone, trismus and fever. Complete blood count (CBC) showed leukocytosis (20,590 cells/mmc) with neutrophilia (17,460 cells/mmc); erythrocyte sedimentation rate (ESR) was slightly higher than normal values and C reactive protein (CRP) values were very high (15.88 mg/dL). The persistent headache alerted physicians and the patient was requested to undergo a computed tomography angiography (CTA) of the head and neck, which showed a right para- and retropharyngeal abscess, spreading in the right side of oral floor and in submandibular space. The abscess eroded the wall of right jugular vein, causing complete thrombosis of the vessel extended to the right sigmoid sinus. There was no evidence of brain lesions. Thrombophilic panel did not show abnormalities. The patient was treated with warfarin 5mg and intravenous antibiotic therapy (meropenem 2g tid and clindamycin 600 mg tid).
Ten days after admission the patient had complete regression of symptoms. CRP levels decreased (0.42 mg/dL) and leukocytosis became less marked (12,000 cells/mmc).
Three months after admission a follow-up CTA was performed. The abscess was completely disappeared, but there was persistence of the thrombotic process of the right jugular vein and sigmoid sinus.









1-2 CTA at admission demonstrating right oropharyngeal abscess and jugular vein thrombosis, with venous air embolism
3-4 3 months CTA follow-up demonstrating disappeareance of abscess but persistence of jugular vein thrombosis

DISCUSSION

Despite the increasing interest on this syndrome, it does not seem that the incidence of Lemierre's syndrome is increasing. [3] However, in the literature only a small number of cases of this disease due to an infection in the dental region are reported.
In our case the syndrome was incomplete, because we did not demonstrate septic dissemination from the infective focus.
Use of anticoagulation in this syndrome is controversial and there is not strong evidence of positive effects of anticoagulants in the literature. [1-3] Some cases, in which there is an involvement of cerebral venous sinuses, may require a more aggressive treatment.[3] In this case, considering also the young age of the patient and lack of comorbidity, we decided to have an aggressive therapeutic approach. After three months the abscess disappeared, but there was no recanalization of the jugular vein and sigmoid sinus.
Lemierre's syndrome can be a sneaky disease, with no pathognomonic symptoms and signs and should be excluded in all cases of jugular vein and venous sinuses thrombosis.

[1] Karkos PD, Asrani S, Karkos CD et al. Lemierre's Syndrome: a systematic review. 2009 Laryngoscope. 119:1552-9
[2] Nov D, Rachmiel A, Levy-Faber D and Emodi O. Lemierre's syndrome from odontogenic infection: review of the literature and case description. 2015. Ann Maxillofac. Surg. 5:219-25
[3] Johannesen KM and Bodtger U. Lemierre's syndrome: current perspectives on diagnosis and management. 2016. Infect. Drug. Resist. 9:221-7

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