### Case Report

#### J Chin Med Assoc

2004:67:419-421

Yen-Jun Lai<sup>1</sup> Jiing-Feng Lirng<sup>1,2</sup> Feng-Chi Chang<sup>1</sup> Chao-Bao Luo<sup>1</sup> Michael Mu-Huo Teng<sup>1,2</sup> Cheng-Yen Chang<sup>1</sup>

# <sup>1</sup> Department of Radiology, Taipei Veterans

### **Key Words**

lemierre syndrome; pharyngitis; veins jugular; veins thrombosis

## **Computed Tomographic Findings in Lemierre Syndrome**

Lemierre syndrome, also known as postanginal septicemia, has long been taken as an uncommon condition with life-threatening potential. It, resulting from acute oropharyngeal infection, could lead to septic thrombophlebitis of the internal jugular vein (IJV). Substantial decrease of mortality and morbidity with the introduction of antibiotics has made this syndrome into a status that is frequently forgotten or overlooked when it appears. The purpose of this article is to refresh awareness on the side of physicians of this syndrome by means of modern image tools. As such, even this dire neck infection is rare after the advent of sophisticated antibiotics, its unique computed tomography findings can facilitate the correct image diagnosis and pertinent treatment.

emierre syndrome, known as postanginal sepsis or Inecrobacillosis, is a suppurative thrombophlebitis of the IJV directly relative to the acute infections of pharyngeal, periorbital, dental or mastoidal region. It was well depicted by Dr André Lemierre (1875-1956) in 1936, with a morality rate of 90% reported in that preantibiotic era. 1 Clinical manifestations may include sore throat or especially a tonsillar abscess, high fever followed by rigor, and painful neck swelling. Fusobacterium necrophorum, a Gram-negative, anaerobic normal flora of the oropharynx and female genital tract, has been verified as the most common causative agent, followed in order by Enterococcus sp and Streptococcus oralis.<sup>2</sup> Lemierre syndrome is formidable as proceeding to septicemia and septic embolism. The initial mucosa-based infection of the upper aerodigestive tract, even being largely restrained by the empirical antibiotics before the causal organism is identified, can still progress to the trasnfascial neck infection with venous occlusion. Image study, especially Computed tomography (CT) study, has

a definitive role in making the timely diagnosis.

#### CASE REPORT

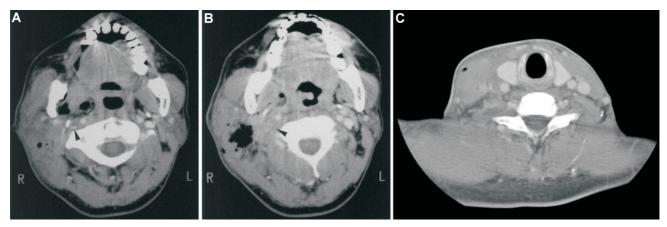
A 51-year-old man was admitted to the hospital for a 10-day painful neck mass followed by 2-day fever and chills. The history can be traced back to his exodontias, treated by a local dentist, the sore throat, and then the painful swelling of neck mass. The patient was a victim of diabetes with poor medication control but denied the consumption of alcohol, tobacco or betel nuts. The physical examination done on arrival showed a patient afebrile with body temperature of 36.8 °C. There was tender swelling of right posterior cervical region, measuring approximately 10 × 10 cm, with moderate pharyngeal erythma. Laboratory data disclosed leukocytosis of 22,300/mm<sup>3</sup>, serum glucose level of 554 mg/dL and high C-reactive protein level of 35.6 ng/mL. The result of the initial blood culture

Received: July 24, 2003. Accepted: February 2, 2004. Correspondence to: Jiing-Feng Lirng, MD, Department of Radiology, Taipei Veterans General Hospital, 201, Sec. 2, Shih-Pai Road, Taipei 112, Taiwan.

Fax: +886-2-2875-7348; E-mail: jflirng@vghtpe.gov.tw

General Hospital; and

<sup>&</sup>lt;sup>2</sup> National Yang-Ming University School of Medicine, Taipei, Taiwan, R.O.C.



**Fig. 1.** A series of contrast enhanced axial computed tomography (CT) of the neck: (**A**) At the level of the oropharynx, showing a gas-containing mass occupying the right-sided masticator and parotid space, spreading to adjacent prestyloid parapharyngeal and carotid space. Almost obliteration of the ipsilateral internal jugular vein (IJV) (arrowhead) due to the intraluminal thrombus is demonstrated. (**B**) On the consecutive caudal cut of image, the IJV (arrowhead) was seen totally occluded, and filled with a hypoattenuating intraluminal thrombus. (**C**) Tissue swelling of the right-sided lateral cervical space is still presented and IJV was recanalized, showing a narrowed, crescent shape lumen (arrowhead).

was negative. CT scan showed a gas-containing mass about  $5 \times 5 \times 10$  cm in the right upper neck from oropharyngeal to supraglottic area (Fig. 1A to 1C). The mass also continuously invaded the adjacent compartments including the masticator space, parotid space, prestyloid parapharyngeal and carotid space with thrombosis of right IJV, evidenced by non-enhanced soft tissue inside the venous channel. A chest X-ray was unremarkable. The patient was given intravenous antibiotics and underwent cervical incision and drainage. At surgery, a huge right abscess formation was found associated with swelling of the subcutaneous tissue and maceration of right carotid sheath. The wound closure was done 7 days later, but due to poor wound healing and pus formation, he received wound debridement again in 8 days, followed by wound closure in another 20 days. The tender mass in the neck improved. The patient was discharged on oral cefadrocil, hypoglycemic agents, and an anti-inflammatory drug to achieve subsequent complete recovery.

### **DISCUSSION**

Lemierre syndrome, septic thrombophlebitis of the IJV, is a complication of an oropharyngeal infection, commonly acute pharyngitis or tonsillitis. According to

a series review by Sinave and colleagues, Fusobacterium necrophorum, an obligate Gram-negative anaerobic bacillus, is the most frequently isolated organism for causing Lemierre syndrome, accounting for 81% of cases, followed by other Fusobacterium species, such as Fusobacterium nucleatum for 11%, and other Gram negative organisms like Enterococcus spp. for 8%. Of 12.8% of documented cased, the culture is inconclusive.

The highest incidence of oropharynx is palatine tonsils (87.1%), while the rest is secondary to odontogenic infection or undetermined.<sup>4</sup> The invasion of the lateral pharyngeal space and IJV thrombophlebitis is usually due to direct extension, or lymphatic or venous dissemination.<sup>4,5,6</sup> Once IVJ is thrombosed, the hematogenous spread can lead to septicemia and distal embolization, by which lung is the most common target.<sup>4</sup>

Many patients with Lamierre syndrome initially present with sore throat and fever only, with more than 50% of patients suffering from swollen and/or tender neck.<sup>3,4</sup> On the other hand, some patients present with pulmonary infection, preceding the overt neck syndromes. Although most patients reported are healthy teenagers or young adults, without apparent medical history,<sup>3,4,7</sup> infants and aging patients with or without underlying chronic disease are not the exception.<sup>4</sup>

The integral parts of Lemierre syndrome consist of oropharyngeal infection and IJV thrombosis. 3,7,8 How-

ever, its critical feature is the occurrence of metastatic thromboemboli, especially septic pulmonary emboli, which usually develops rapidly and is life-threatening.<sup>4,6</sup> CT of the neck with contrast enhancement is the imaging modality of choice in demonstrating the thick enhancing wall and the non-enhanced filling defect of the IJV along with swelling of the surrounding structures. The clinicians should be vigilant of the development of the various manifestations of pleuropulmonary disease, like lobe consolidation, pleural effusion, peripheral pulmonary nodules and even pulmonary abscess formation or empyema.<sup>5</sup> On the other hand, the main role of radiologist is to award the clinician of this innocent-appearing pharyngitis at the early stage before the more devastating condition occurs. Treatment for Lemierre syndrome involves prolonged high-dose intravenous antibiotics against Fusobacterium necrophorum with good anaerobic coverage. Surgical drainage of any abscess that develops is recommended. However, ligation of the internal jugular vein or anticoagulation remains controversial and is typically reserved for cases that are refractory to treatment with antibiotics alone.7,11

Although quite rare nowadays, Lemierre syndrome still poses a life-threatening complication that easily deceives itself in our daily practice. Early diagnosis can expedite the treatment and decrease the unwanted complications. Therefore, CT absolutely stand high as the key modality in being reminiscent of the forgotten disease. Once the diagnosis of Lemierre syndrome is suspected, antibiotics with good anaerobic activity should be included before obtaining the culture result.

#### REFERENCES

- 1. Lemierre A. On certain septicaemias due to anaerobic organism. *Lancet* 1936;1:701-3.
- Sinave CP, Hardy GJ, Fardy PW. The Lemierre syndrome: suppurative thrombophlebitis of the internal jugular vein secondary to oropharyngeal infection. *Medicine* 1989;68:85-94.
- 3. Screaton NJ, Ravenel JG, Lehner PJ, Heitzman ER, Flower CDR. Lemierre syndrome: forgotten but not extinct-Report of 4 cases. *Radiology* 1999;213:369-74.
- 4. Chirinos JA, Lightsten DM, Garcia J, Tamariz LJ. The evolution of Lemierre syndrome: Report of 2 cases and review of the literature. *Medicine* 2002;81:458-65.
- Nguyen-Dinh KV, Marsot-Dupuch K, Portier F, Lamblin B, Lasjaunias P. Lemierre syndrome: Usefulness of CT in detection of extensive occult thrombophlebitis. *J Neuroradiol* 2002;29:132-5.
- Blomquist IK, Bayer AS. Life-threatening deep fascial space infections of the head and neck. *Infect Dis Clin North Am* 1988;2:237-4.
- 7. Gong J, Garcia J. Case report: Lemierre syndrome. *Eur Radiol* 1999;9:672-4.
- 8. Holland BW, McGuirt WF Jr. Imaging quiz case 2. Lemiere syndrome: Septic thrombophlebitis of the internal jugular vein, or "postanginal" sepsis. *Arch Otolaryngeal Head Neck Surg* 2000;126:1500-4.
- 9. Albertyn LE, Alcock MK. Diagnosis of internal jugular vein thrombosis. *Radiology* 1987;162:505-8.
- 10. Ostrum DS. Thrombosis of the internal jugular vein. Findings on MR and color Doppler imaging. *AJR* 1991;157:1358-9.
- 11. Alvarez A, Schreiber JR. Lemierre syndrome in adolescent children: anaerobic sepsis with internal jugular vein thrombophlebitis following pharyngitis. *Pediatrics* 1995;96:354-9.