CASE REPORT

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Lemierre's Syndrome Originated from the Odontogenic Infection: A Case Report

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Also called necrobacillosis or postanginal sepsis, Lemierre's syndrome (LS) is an uncommon but potentially lethal complication of odontogenic infection. A 27-year-old male diagnosed with Ludwig's angina was transferred from a local hospital due to continuous fever and chills after incision and drainage under general anesthesia. The swelling of both submental and submandibular area subsided, but the fever and chills persisted. While generalized malaise improved, sepsis developed together with the deterioration of liver function. The chest computed tomography scan revealed multiple cavitations throughout both lungs, which were diagnosed as septic pulmonary embolism. After consulting the department of infectious diseases, the patient was treated with intravenous antibiotics focusing on vancomycin and additional antibiotics. After 3 weeks of treatment, the patient recovered completely. Despite its decreased mortality, dentists are not familiar with LS, and it is difficult to diagnose correctly. In this paper, we report a case and present a review of literature.

Key Words: Ludwig's angina; Odontogenic infection; Pulmonary embolism

Introduction

Also called necrobacillosis or postanginal sepsis, Lemierre's syndrome (LS) is characterized by acute oropharyngeal infection complicated by internal jugular venous thrombosis (IJVT) secondary to septic thrombophlebitis and by metastatic infections affecting various distant organs, most commonly the lungs[1]. LS was a life-threatening syndrome before antimicrobial agents became available. With the introduction of antibiotics, however, the incidence and mortality decreased. That is why LS is known as the "forgotten disease"[2].

We report an unusual case of LS from primary odontogenic infection and review the published literature. This case could be diagnosed and treated as LS with consultation with the Department of Infectious Diseases since the patient's generalized

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fever was not controlled in spite of proper surgical intervention and long-term antibiotic therapy.

Case Report

A 27-year-old male diagnosed with Ludwig's angina was transferred from a local hospital due to continuous generalized fever and chills after incision and drainage of pus under general anesthesia. The infection origin was estimated to be a root rest of the left mandibular first molar and chronic periodontitis (Fig. 1). Pus was discharged, and swelling of both submandibular and submental area subsided (Fig. 2). The generalized fever and chills persisted, however. While generalized malaise improved, sepsis developed together with the deterioration of liver function.

Fig. 1. Panoramic view exhibiting the root rest of the left mandibular first molar and chronic periodontitis.

Laboratory tests revealed increased white blood cell count and serum C-reactive protein (Table 1). The neck doppler ultrasonography showed no evidence of bilateral IJVT. The computed tomography of the chest showed bilateral multiple scattered nodular lesions of varying sizes as strongly suggested in

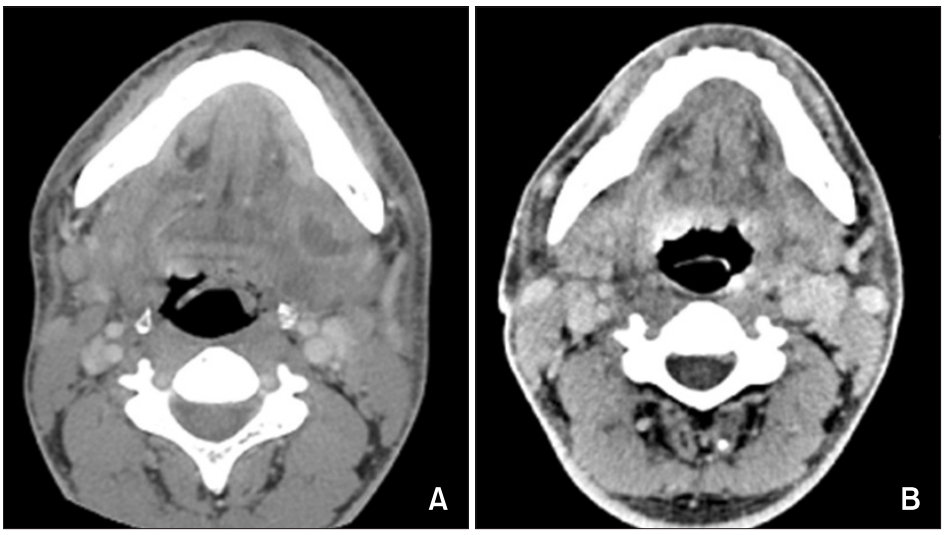

Fig. 2. Contrast-enhanced computed tomography of the neck before surgical intervention (A) and after surgical intervention for Ludwig's angina (B).

Table 1. Laboratory findings in this case

	Normal	HD#1	HD#3	HD#5	HD#7	HD#9	HD#15
WBC (x10^3/mm^3)	4~10	13.3	19.7	18.0	13.9	10.5	6.1
Hct (g/dl)	39~52	42.4	40.8	35.3	35.3	36.2	36.3
PLT (x10^3/mm^3)	150~450	166	142	207	245	302	231
Seg (%)	50~75	88.9	85.2	86.0	75.8	72.7	66.0
PT (sec)	9.0~13.2	17.8	18.7	14.0	13.4	12.8	9.4
sCRP (mg/dl)	0~0.3	3.7	5.2	4.8	3.0	2.1	1.1

HD: hospitalization day, WBC: white blood cell count, Hct: hematocrit count, PLT: platelet count, Seg: segmented neutrophil count, PT: prothrombin time, sCRP: serum C-reactive protein.

Table 2. Reviews of Lemierre's syndrome cases especially with odontogenic source

	Source	Pathogens	IJVT	Septic emboli	Antibiotics
Le Moal et al.[9]	Periodontal disease	*Fusobacterium necrophorum*	(-)	Spine	Penicillin clindamycin
Sonsale et al.[10]	Dental abscess	*Fusobacterium necrophorum*	(-)	Knee	Metronidazole
Shibasaki Warabi et al.[11]	Caries	Unknown	(+)	Lungs, brain	Penicillin metronidazole
Duquesne et al.[12]	Gingivitis	*Fusobacterium necrophorum*, *Staphylococcus aureus*, and *Enotrophomonas xanthomonas*	(-)	Meningitis	Cefotaxime vancomycin amikacin
Tan et al.[13]	Caries and periodontitis	*Fusobacterium necrophorum*	(+)	Lungs, meningitis	Penicillin metronidazole
Juárez et al.[14]	Dental abscess	*Streptococcus intermedius* and *Bacteroides fragilis*	(-)	Lungs	Levofloxacin vancomycin

IJVT: internal jugular vein thrombosis.

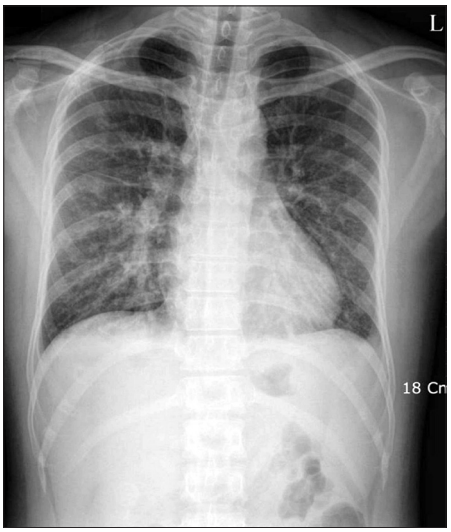

Fig. 3. The plain chest radiograph reveals multifocal cavitations (septic emboli) throughout both lungs.

Fig. 4. The chest computed tomography scan reveals septic emboli throughout both lungs.

the plain chest radiograph (Figs. 3 and 4). After consulting the Department of Infectious Diseases, the diagnosis of LS was confirmed, and the patient was treated with intravenous antibiotics focusing on vancomycin and additional antibiotics such as augmentin and ceftriaxone. With the improvement of the liver function, his infection symptoms including generalized fever and malaise disappeared. After 3 weeks of treatment, the patient recovered completely, and he was discharged.

Discussion

First described by Dr. Andre Lemierre in 1936, LS is an uncommon but potentially lethal complication that affects young, healthy people. The incidence rates are between 0.6 and 2.3 for every million people. It is found more commonly among males, with male to female ratio of approximately 2 : 1[3]. LS is a severe complication of head and neck infections, resulting in lateral pharyngeal space infection and IJVT followed by septicaemia and metastatic infections predominantly in the lungs; the liver, spleen, kidney, meninges, heart, muscle, and skin can also be involved[2].

An extensive search of English literature from

2001 to 2008 was undertaken by Rosado et al.[4] to identify published cases, using multiple search criteria such as LS, *Fusobacterium necrophorum*, which is known as the most common bacterium causing LS, postanginal sepsis, and IJVT. A total of 99 cases of LS were found and reviewed. Based on their review, the primary site of infection was oropharyngeal infection (60% of the cases), followed by mastoiditis (15%). They found only 7 cases with primary odontogenic infection (7%), most of which had developed from gingivitis and periodontitis as well as dental caries. Only 2 cases had the IJVT (Table 2). Five blood cultures were positive for *F. necrophorum*, which plays a minimal role in LS from odontogenic infection. Most of the patients, despite the initial treatment focusing on *F. necrophorum*, continued to suffer from fever and chills that precede septicaemia. Mononucleosis was associated with 12% of the cases; therefore, mononucleosis has been considered a predisposing factor to invasive LS. Although magnetic resonance (MR) has been introduced, recent literature suggests that color doppler ultrasonography and high-resolution CT are still important imaging tools in identifying IJVT and pulmonary septic emboli or metastases, which are critical in the spread of the disease[4]. A plain chest radiograph may not be effective in the early diagnosis of septic emboli[5].

The pathogenesis of LS involves the development of IJVT on one of the branches of internal jugular vein caused by focal sepsis mostly localized in the oropharynx, leading to generalized multi-organ metastatic infections[2]. Frequent symptoms include sore throat, high-temperature fever and chills, and painful swelling in the neck, typically along the anterior border of the sternocleidomastoid muscle (SCM). Edema and pain in the mandible angle or located anterior and parallel to SCM reflect the development of IJVT, which occurs in 26~45% of all the cases. Dysphagia and trismus may be present as a result of infection of the lateral pharyngeal space[6]. In our case, the patient had fever refractory to conventional treatment, edema and pain in both mandible angle, and dysphagia, which progressed to Ludwig's angina from odontogenic source.

In most cases, *F. necrophorum* is the tentative causative microorganism[4]. Other pathogens that can cause LS include other *Fusobacterium* species, *Bacteroides* species, and groups A, B, and C *Streptococcus*. A polymicrobial bacteraemia is present in 10~30% of cases[7]. It is difficult to identify solely the *Fusobacterium* species, which is often found in association with other pathogens[8]. In the case reported here, *F. necrophorum* was not identified in the blood culture.

Pulmonary involvement in LS is extremely common (seen in as much as 97% of the cases). The metastatic foci of infection in the lungs can be manifested by dyspnea, pleuritic chest pain, or hemoptysis[2]. The patient in this case had pulmonary lesions that were found based on the plain chest radiograph and confirmed in the chest CT. Ultrasound or tomographic scans are used to confirm IJVT. The patient in our case had neither respiratory manifestations nor IJVT as confirmed by neck doppler ultrasonography but showed leukocytosis combined with neutrophilia. Deterioration of the liver function was found in 50% of the cases, similar to our case.

In summary, the followings are typical findings suggestive of LS:

1. History of acute oropharyngeal infection about one week prior to the manifestation of symptoms
2. Pain and swelling in the cervical region
3. High fever and chills
4. Pulmonary involvement
5. IJVT
6. Metastatic infections at other sites
7. Anaerobic result in blood cultures or cultures obtained from other sites of metastatic infection

The patient in our report showed 1, 2, 3, 6, and 7.

Given the severity of this disease, and because its correct diagnosis is difficult, familiarity with all aspects of the disease is essential. This syndrome

needs immediate diagnosis and treatment with the assistance of the Department of Infectious Diseases. Early recognition and high-dose antibiotics are critical elements in reducing mortality. Dentists must be alert in detecting these rare cases with odontogenic origin.

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