Review Article

Lemierre's syndrome from odontogenic infection: Review of the literature and case description

Access this article online
Website: www.amsjournal.com
DOI: 10.4103/2231-0746.175746
Quick Response Code:

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ABSTRACT

Lemierre's syndrome (LS) is a rare potentially fatal sequel of head and neck infection, classically described as thrombophlebitis of the internal jugular vein (IJV) with cervical space infection extending into the thorax. Our objective was to answer the clinical question: "Does Lemierre syndrome (LS) from odontogenic infection differ from nonodontogenic LS in regard to clinical sequence, treatment, and survival." We reviewed the literature on the management of LS over the last two decades, with a focus on LS from odontogenic infection. Such a case is presented in order to portray the clinical sequence. Only 10 cases met the inclusion criteria (including the case presented). The recorded data were analyzed in comparison to large case series reviewing LS. Our data reflect the moderate differences in regard to IJV thrombosis and bacteriogram. There is an overall rise in published LS cases in the last 20 years. Odontogenic infection leading to LS is scarce, yet with survival rates similar to nonodontogenic LS. Repeated surgical interventions and aggressive wide spectrum antibiotic therapy remain the treatment of choice.

Keywords: Descending mediastinitis, Lemierre's syndrome, necrotizing fasciitis, odontogenic infection, postanginal septicemia

INTRODUCTION

Lemierre syndrome (LS), also termed postanginal septicemia and human necrobacillosis, was first described in 1936 by Lemierre as an oropharyngeal infection, followed by septic thrombophlebitis of the internal jugular vein (IJV) and subsequent hematogenic spread of the infection, via septic emboli.[1]

In Lemierre's original publication in Lancet, out of 20 cases, 18 had died.[1] Indeed, before the antibiotic era, the mortality was 90%. With the introduction of antibiotics, the incidence and mortality of LS decreased to such a degree that it was termed "the forgotten disease."[2,3]

LS mostly affects healthy young adults. 70% of people with LS are 16–25 years of age. LS occurs in about one per million people, with a mortality rate approximately 4–12%.[4]

The infectious causative foci lie in the head and neck region, mostly pharyngitis or tonsillitis accounting for over 85% of LS cases.[5]

Mastoiditis and odontogenic infections are far less common, accounting for 3% and 2% of LS cases, respectively, as the site of primary infection. Cases resulting from central venous catheterization and cervical intravenous (IV) drug abuse have also been reported.[4]

The clinical symptoms may differ depending on the primary infection site, but are almost always accompanied with local pain and sore throat (over 80% of cases), fever, odynophagia, trismus, and nausea.[6]

Cite this article as: Noy D, Rachmiel A, Levy-Faber D, Emodi O. Lemierre's syndrome from odontogenic infection: Review of the literature and case description. Ann Maxillofac Surg 2015;5:219-25.

The purpose of this paper is to review the literature on epidemiology and clinical management of such cases over the last two decades, with a unique focus on LS secondary to odontogenic infection. Also, such a case is presented in order to portray the clinical sequence.

CASE DESCRIPTION

An otherwise healthy 30-year-old male sailor from Ukraine presented himself at the emergency room in our institution, with a history of a 5-day toothache and dysphagia to solids with fevers up to 40°C. On physical examination, the patient was febrile at 38.7°C, with tachypnea, and tachycardia. He had a left submandibular tender swelling with erythema and cellulitis, with spontaneous pus discharge. Interincisal opening was within normal range; some left mandibular teeth (2nd premolar and wisdom tooth) were decayed with minimal vestibular fluctuation. The white blood cell (WBC) count was 14.10 × 10^3/mL with a left shift of 92%. A computerized tomography (CT) demonstrated diffused edema of the epidermal tissues lateral to the left ramus and body of the mandible down to the base of the neck with a localized collection measuring 4 cm. In addition, the left masseter was edematous, and signs of involvement of para-pharyngeal space were suspected. There was no bony involvement.

The patient was rushed to the operating room where, under general anesthesia, he underwent left mandibular teeth extractions and transcervical productive drainage of the submandibular abscess with local debridement of necrotic subcutaneous tissue and placement of sheet drains. Tissue culture and biopsy were also taken. He was consequently started on empiric IV antibiotics clindamycin 300 mg × 4/day.

In the next 3 postoperative days the patient demonstrated relative improvement; a decrease in submandibular swelling and WBC count (7.36 × 10^3/mL), but spiking fevers up to 39.7°C persisted. On the 4th postoperative day, the patient became tachypneic and tachycardic, with bilateral pleuritic chest pain and dyspnea with leukocytosis (17.16 × 10^3/mL) and oropharyngeal candidiasis. Consequently, the IV antibiotic dosage was changed to clindamycin 900 mg × 3/day and ciprofloxacin 400 mg × 2/day. On the 6th postoperative day a repeat contrast-enhanced CT demonstrated the collections in the left neck base with involvement of the sterno-cleido-mastoid muscle with suspected thrombosed left jugular vein [Figure 1], mild pericardial effusion and bilateral loculated pleural effusion. Small vessels in lung fields were suspected as septic pulmonary emboli (SPE). In addition, mediastinal lymphadenopathy was noted with hepatomegaly.

Antibiotic regimen was replaced based on the sensitivity tests with wide spectrum antibiotics IV vancomycin 1 g × 2/day and metronidazole 500 mg × 3/day together with antifungal fluconazole 150 mg × 1/day.

Transthoracic echocardiogram (TTE) demonstrated the pericardial effusion but no sign of tamponade or vegetations.

A workup for tuberculosis, bartonnela, and an immunocompromised status was negative, and liver functions were within the normal range.

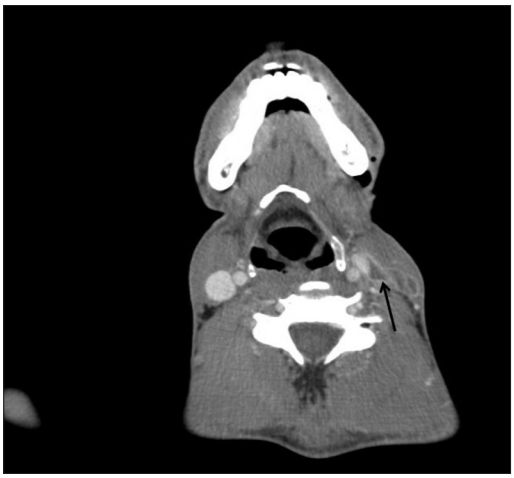

Figure 1: Axial contrast-enhanced computerized tomography at the level of the hyoid bone showing filling defect/thrombus in situ within the left jugular vein (arrowhead)

WBC count rose to 40.2 × 10^3/mL.

Bilateral chest tubes were productive with pus-like pleural effusion.

On the 8th postoperative day, a third contrast-enhanced CT demonstrated bilateral (BLT) loculated pleural effusion with pericardial effusion [Figure 2]. Status postdrainage of the cervical abscess with collection reaching through the vascular compartment, on the left side of the mediastinum and retrosternum to the level of the xyphoid, with the involvement of the subdermoid fat in the left lower neck containing air bubbles [Figure 3].

Hemoculture was positive for *Staphylococcus capitis* and alpha hemolytic streptococci.

The patient's skin in the anterior upper left thorax demonstrated a violacious hue, with "crushing snow" sound on palpation [Figure 3].

The patient was rushed to the operating room where he underwent left open fasciotomy and consequently bilateral thoracoscopy with lung decortication [Figures 4-6]. The patient was transferred to the Intensive Care Unit (ICU) where he remained for 3 days. IV antibiotic regimen was changed to imipenem 500 mg × 4/day and vancomycin 1 g × 1/day, and preventive anticoagulatory enoxaparin sodium (clexane 40 mg × 1/day) was started.

In the following days, the patient's condition was steadily improved; however, an ultrasound (US) guided chest drain was inserted due to a left extrapleural collection. Edema was noted in the patient's left hand due to a basilic vein thrombosis.

A second TTE demonstrated an unchanged pericardial effusion, and still no sign of tamponade or vegetations.

At this point, the patient was afebrile. His pulmonary, hemodynamic, and radiographic condition normalized. The operative skin wounds showed healthy granulation tissue.

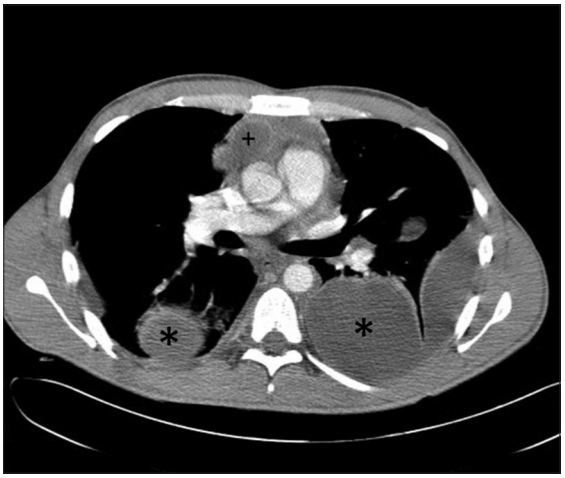

Figure 2: Axial contrast-enhanced computerized tomography at the level of the upper mediastinum showing bilateral loculated lung abscesses (*) and collection within the mediastinum (+)

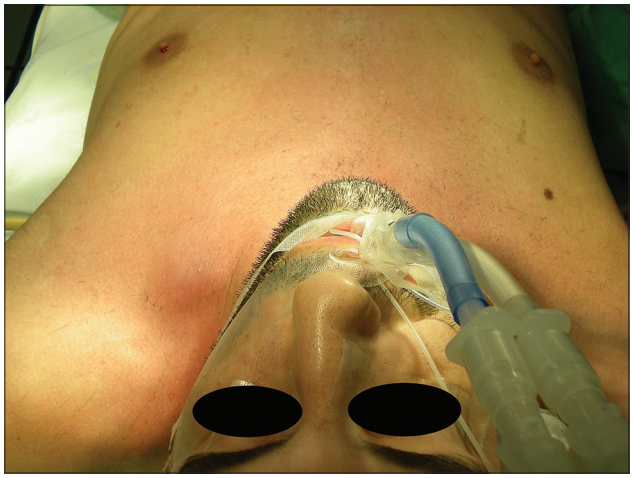

Figure 4: A Supine photograph was taken just before the cervical fasciotomy and thoracoscopy. A violaceous hue in the left lower neck and upper thorax is noticeable

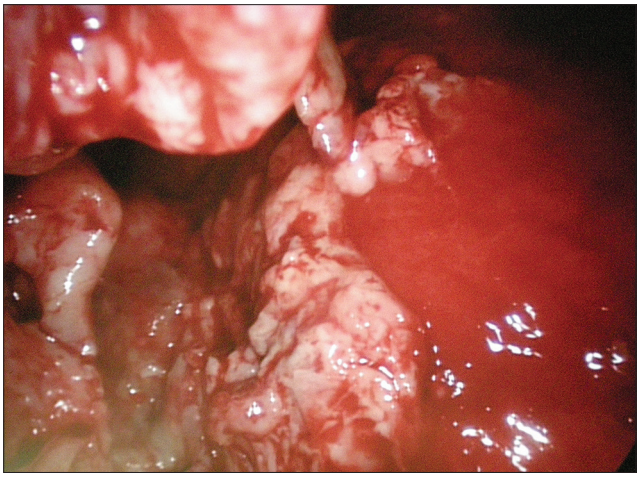

Figure 6: An intraoperative photograph taken the during lung decortication. Pus and fibrin clots covering lung tissue were apparent bilaterally

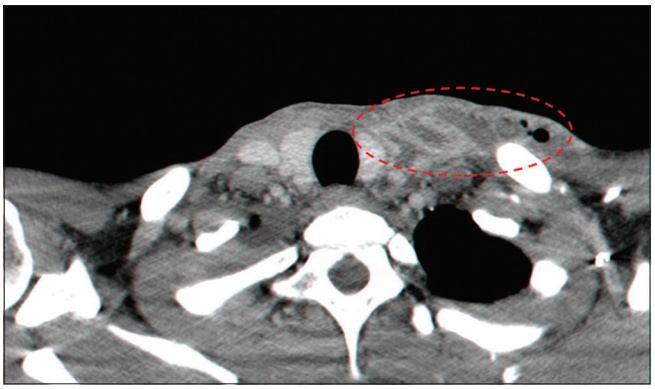

Figure 3: Axial contrast-enhanced computerized tomography at the level of the clavicles showing air bubbles and collection foci in the left anterior part of the lower neck (circumscribed)

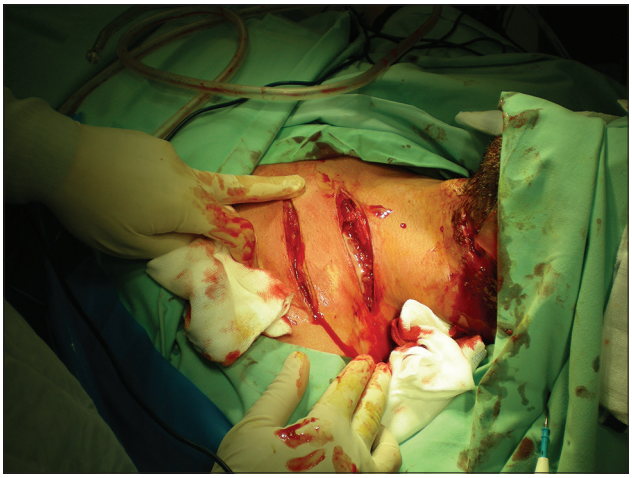

Figure 5: Intraoperative transverse cuts in the left lower neck and upper thorax with pus discharge

He was transferred in good general condition to another institution after a total of 24 days of hospitalization.

DISCUSSION

We performed a search confined to the English scientific literature in PubMed database, trying to identify the papers describing LS secondary to an odontogenic infection in the last two decades. The rationale for this time frame was the development of diagnostic imaging techniques, namely CT and magnetic resonance imaging (MRI). Comparison of earlier cases with recent ones may be somewhat anachronistic in regard to the amount of data gathered, especially pertaining IJV thrombophlebitis and SPE.

Authors vary as to which the diagnostic criteria are essential to define LS. We used the following combination to define LS:[7]

- Oropharyngeal illness in the preceding 4 weeks
- Evidence of metastatic lesions in lungs and/or another remote site
- Evidence of IJV thrombophlebitis or the isolation of *Fusobacterium necrophorum* or *Fusobacterium* sp. from the blood cultures of a normally sterile site.

Due to the retrospective nature of this study, it was granted an exemption in writing by Rambam Health Care Center's Institutional Review Board.

Our report reviews 10 cases of LS due to odontogenic infection published in the last two decades [Table 1]. Perhaps the most recent extensive reviews on the subject of LS are Riordan's, reviewing 222 cases from 1970 to 2007,[7] and Karkos *et al*., reviewing 114 cases from 1950 to 2007.[19]

The fact that these large case series contain 1–2% cases of LS due to odontogenic infection is negligible. The small size of the study group (10 cases) may impair validity. However comparison to larger case series may outline the uniqueness of LS due to odontogenic infection [Tables 2 and 3].

The most common presenting symptoms of LS are fever, chills, and rigors together with stiffness of the neck. A sore throat with dysphagia and disproportionate local pain with or without cellulitis which may progress to trismus and restricted mouth opening are also common mainly in conjunction with deep neck space infection. Initial lab studies suggest sepsis. Also, limitation of head movement, pleuritic chest pains, hemoptysis, tachycardia, and dyspnea may be present. The infection affects the deep spaces of the carotid sheath and erodes the IJV.[20] A possible explanation for the high IJV thrombophlebitis we found in the setting of odontogenic LS may be the plethora of bacteria found in the oral cavity together with the lack of adequate imaging techniques in chronologically older cases. Septic emboli from the thrombosed jugular vein may disseminate hematogenously throughout the body, mainly to the lungs, creating septic lacunae, infarcts, and abscesses.[21]

Table 1: Lemierre syndrome due to odontogenic infection - literature review

Investigator	Presenting symptoms	Onset	Dental origin	Metastatic lesions	Jugular vein thrombophlebitis	Bacterial profile
Noy, 2014	Spiking fevers, tachypnea, pleuritic chest pain	3 days	blt. mandibular molars	blt. Pleural effusions, mediastinitis,	+	alpha-hemolytic *Streptococci*
Noy, 2013	Spiking fevers, tachypnea, tachycardia, lt. submandibular cellulitis with spontaneous pus discharge	5 days	Carious lt. mandibular molars	blt. pleural effusions, pericardial effusion, lt. basilar vein thrombosis	+	alpha-hemolytic *Streptococci, Staphilococcus capitis*
Rosado, 2009[8]	Fever, dyspnea, submandibular cellulitis, sublingual swelling	3 days	rt. mandibular 3rd molar	Pleural effusions, SPE	+	*Streptococcus Salivarius*
Malis, 2008[4]	Spiking fever, restricted mouth opening, dysphagia, lt. submandibular swelling extending to neck, rt. deviation of uvula, blt. pleuritic chest pains	5 days	Total wisdom teeth extraction	Pericardial effusion, blt. pleural effusions	+	alpha-hemolytic *Streptococci*
Juarez Escalona, 2007[9]	Persistent fever, rt. submandibular and sublingual swelling	5 days	rt. mandibular 3rd molar	SPE	+	*Streptococcus Intermedius, Bacteroides Fragilis*
Duquesne, 2007[10]	Fever, pulsatile fontanele, GCS = 8	8 days	Gingiva	Trombophlebitis of rt. petrosal sinus	None	FN
Shibasaki Warabi, 2005[11]	rt. temporal pain, fever, chills, lt. "cord" sign, rt. Hemiparesis	7 days	rt. maxillary abcess, caries	Lung abcesses, cerebreal infarctions and emboli	+	n/a
Sonsale, 2004[6]	Fever, malaise, swelling of rt. Knee	3 weeks	Dental abcess	isolated septic knee	None	FN
Tan, 2003[12]	Spiking fevers, mild neck stiffness, throbbing headache	3 weeks	Maxillary and mandibular carious molars	SPE, bacterial meningitis	+	*Fusobacterium Nucleatum*
Carlson, 1994[13]	Malaise, myalgia, fever and chills, vomiting, lt. neck pain	6 days	Multiple severely carious teeth	SPE	+	FN
			Sub total: 10 cases			
Ruskin, 2009[14]	lt. submandibular swelling with green discharge and blt. Proptosis	2 weeks	lt. mandibular 3rd molar extraction	None	+	*Streptococci, Staphilococcus Epidermidis*
Le Moal, 2005[15]	Febrile, low back pain	3 months	Periodontal disease mandibular molar. extracted	Isolated vertebral osteomyelitis	None	FN
	Backache, intermittent fever, sweats	n/a	n/a	Isolated vertebral osteomyelitis	None	FN
	Backache, fever, sweats	n/a	Generalized periodontal disease	Isolated vertebral osteomyelitis	None	*Fusobacterium Nucleatum*
Cook, 2005[16]	n/a	n/a	maxillary, mandibular	SPE	None	n/a
Esposito, 2004[17]	Headache, rt. neck and rt. mandible pain	n/a	rt. mandibular 2nd molar with perapical abcess	n/a	None	FN
Christensen, 1993[18]	lt. pleuritic chest pains, fever	3-4 weeks	n/a	SPE	None	*Streptococcus Intermedius*
			Sub total: 7 cases			
			Total: 17 cases			

rt.=Right; lt.=Left; blt.=Bilateral; SPE=Septic pulmonary emboli; n/a=Not available; FN=Fusobacterium necrophorum. The cases in the upper part side of the table (10 cases) were included as odontogenic LS, whereas those in the lower part (7 cases) were excluded as such

Table 2: Lemierre syndrome - epidemiological considerations. Comparison of the current review with large case series

	Noy, 2014	Riordan, 2007[7]	Karkos, 2009[19]
No. of cases	10	222	114
Time frame	1990-2014	1970-2007	1950-2007
Median age (years)	30	19	22
% male	78%	57-68%	45%
Presenting symptoms	Fever and chills, malaise, neck pain, dysphagia, restricted mouth opening, pleuritic chest pains	Fever, chills, rigors, stiffness of neck, dysphagia, restricted opening	Fever, sore throat, neck pain
Onset - Median value	5 days	5 days	n/a
Imaging modalities	CT-78%, MRI-33%	n/a	CT-55%, MRI-6%
Jugular vein thrombophlebitis	78%	59%	n/a
Bacteriogram	*Fusobacterium spp.* 44%, *alpha hemolytic stretoccoci.* 44%, bacteroides fragilis 11%, staphyloccocus capitis 11%	Fusobacterium spp. 86%, other organism w/o Fusobacteria 8%	Fusobacterium spp. 90%, Other 10%
Metastatic septic lesions	Lungs 78%, septic arthritis (knee) 11%, peripheral veins (basilar) 11%, cranium (petrosal sinus) 11%	Lungs 92%, meningitis 1.4%, septic arthritis (knee) 11%	n/a
Surgical treatment	78% treated surgically, 67% IO, 52% EO, 22% TO	n/a	n/a
Antibiotics	Metronidazole 78%; clindamycin 67%; vancomycin 44%; penicillins 44%; cephalosporins 33%; aminoglicosides 33%; fluoroquinolones 33%	Carbapenem OR metronidazole; carbapenem + metronidazole for oral infection	Penicillin with beta lactamase resistance, clindamycin, metronidazole, chloramphenicol
Anticoagulant therapy	33%	23%	30%
Median hospital stay	21 days	18 days	25 days
Outcome (survival)	90%	95%	95%

All percents relate to the total number of subjects in the relevant series. n/a=Not available; CT=Computed tomography; MRI=Magnetic resonance imaging; IO=Intra oral; EO=Extra oral; TO=Thoracotomy

Table 3: Lemierre syndrome - extrapulmonary morbidity. Comparison of the current review with large case series

	Noy, 2014	Riordan, 2007[7]	Karkos, 2009[19]
Intracranial	22%	16%	30%
Arthritis/osteomyelitis	11%	11%	22%
Deep neck space infection	55%	10%	14%
Cardiac	22%	2%	7%
Liver	n/a	4.5%	6%
Spleen	n/a	3.5%	6%
Lower cranial nerve palsy	11%	3%	3%
Orbital	n/a	n/a	5%
Mediastinitis	30%	2%	n/a
Skin	11%	4%	n/a
Total	33%	27%	n/a

All percents relate to the total number of subjects in the relevant series. n/a=Not available

Pulmonary lesions are the most common metastatic complications of LS. A lethal sequel of LS in the context of this article is descending necrotizing mediastinitis (DNM).[4]

DNM spreads along the deep fasciae of the neck, unlike the hematogenic spread of LS.

DNM has a high mortality rate of over 40%, even with the use of high dose wide spectrum antibiotics.[22] Malis *et al*. reported one case of 23-year-old male with LS secondary to postextraction infection with the sequel of DNM. Alpha hemolytic *Streptococcus* was isolated. Septic emboli were found in the lungs with the involvement of the mediastinum.[4]

In order to assess the extensiveness of LS, we examined the associated extrapulmonary morbidity [Table 3]. LS of odontogenic origin is probably likely to involve deep neck space infections due to the vicinity of primary fascial spaces from the oral region to deep cervical spaces, such as the para-pharyngeal space.[20]

The flora of deep neck space infections comprises aerobic and anerobic bacteria with anerobes predominating. Such is the case for most oral infections. Bacteria most commonly isolated from deep fascial spaces are *Peptostreptococcus* spp., *Fusobacterium* spp. (predominately *F. nucleatum*), *Bacteroides* spp. and also *Staphylococcus aureus*, group A streptococci, and anerobic bacteria of dental origin.[21] *F. necrophorum* is considered the causative microbiological pathogen of LS, although it normally inhabits the oropharynx. It may erode the jugular vein, disseminating septic emboli to the lungs, brain, and other organs, possibly creating abscess lacunae in both lungs.[23] *F. necrophorum* may be cultured within 48 h in fastidious anerobic agar. However, the nonexistence of *F. necrophorum* in blood cultures does not rule out LS, as in the case mentioned in our report. There is inconclusive evidence whether *F. necrophorum* is a sin qua non for LS. This is perhaps because, in some reports, *F. necrophorum* could not be isolated.

These findings suggest that *F. necrophorum* does not play a key role in deep neck space infections such as para-pharyngeal or retropharyngeal abscesses. Riordan postulates that perhaps *F. necrophorum* has been present but has gone undetected.[7]

Intracranial manifestations that may follow LS include meningitis, cerebral abscess, sinovenous thrombosis, and septic emboli. The route of intracranial infection spread is most likely by the retrograde advancement of septic metastases from the IJV via the sinovenous system.[10,13]

Cardiac manifestations of LS include pericardial effusion, pericardial tamponade, and endo/pericarditis. Nevertheless, these are concerned rare.[19]

In many cases, there is no identifiable abnormality on initial films.[24] Contrast-enhanced CT scan reveals earlier evidence of a filling venous defect, namely thrombophlebitis. Further spread of the infection may demonstrate pleural effusion and septic

thromboses in the lungs. Since IJV thrombophlebitis is frequently unapparent clinically, there is considerable dependence on imaging techniques to detect this important feature.[25]

US scan has been used by some to detect jugular vein thrombosis. Although rapid, low cost, and noninvasive technique it cannot image deeper spaces such as beneath the clavicle or mandible. Some studies showed the failure of US to detect IJV thrombosis that was demonstrated by contrast-enhanced CT.[26]

MRI is a good tool in the initial evaluation of neck infections. However, in emergency situations this diagnostic tool is not always available owing to its high cost. MRI is superior to CT in regard to lesion conspicuity and determining the number of anatomic spaces involved, although the extent of the infection may be exaggerated.[27]

Contrast-enhanced CT remains the modality of choice for an early diagnosis of LS.

Treatment must be prompt. Protection of the airway must be the priority. Consequently, tracheostomy should be considered and admittance to an ICU for the first 72 h is advised.

Aggressive surgical exploration and debridement are mandatory. Surgical drainage of the chest via thoracotomy should be considered in life-threatening situations, as we did in the case described here. Necrotizing infections require not only simple drainage of collections but also radical debridement of all affected tissues. The possibility of several consecutive surgical interventions is high. The transcervical incision is recommended to drain the superior mediastinum. Often there is no need to remove skin tissue as in necrotizing fasciitis. Local wound care in the immediate postoperative period should include wet dressing change. Some recommend the use of saline or hydrogen peroxide every few hours after surgical drainage.

In the pre antibiotic era, in the face of dissemination of septic emboli from a thrombosed IJV and mortality of over 90%, drastic measures were attempted including ligation or excision of the IJV. There are no controlled studies that show the benefit of IJV ligation/excision in the treatment of LS.[28] In his case series, Riordan states that since a large number of cases with the proven thrombophlebitis resolve without IJV surgery, it seems unjustifiable.[7] Perhaps, the only indication for IJV ligation/excision is persistent septic emboli or extending thrombi despite aggressive therapy.

Since LS involves IJV thrombosis and abscess lacunae in the lungs, there are penetrance issues in regard to antibiotic treatment. Two to three class drugs are advised because of the mixed nature of the flora of deep neck space infections. Amoxicillin and clavulanic acid or clindamycin, together with metronidazole remain the first line of defense before further wide spectrum antibiotics are used, such as vancomycin or imipenem.[29] Some researchers contemplate that the clindamycin is not susceptible to the "Eagle effect" as is penicillin. Therefore, it is advisable to use clindamycin empirically as the antibiotic of choice in severe infections.[30]

The use of anticoagulation therapy in the context of thrombophlebitis has been controversial, and no controlled studies exist to date, nor indications for initiation of such treatment. Some authors report the defervescence and clinical improvement upon the initiation of heparin therapy. The rationale is the success in the treatment of septic pelvic thrombophlebitis with heparin. The benefit might be implicated in the case of the IJV.[31] Perhaps the fact that *F. necrophorum* may produce hemagglutinin and consequently platelet aggregation and diffuse intravascular coagulation is an indication for anticoagulant therapy.[13] Bondy and Grant concluded, that although the use of anticoagulation therapy in various specialties (otology, obstetrics, gynecology, and internal medicine) is common, its role in LS is unclear.[32] Anticoagulation had been used more frequently when thrombosis involved the sigmoid or cavernous sinus.[7] The drug of choice was heparin and sometimes low molecular heparin for long-term treatment. The lack of controlled trials in the matter is probably due to the low incidence of LS. Anticoagulation therapy, to accelerate the thrombus breakdown remains a matter of controversy. Albeit, it does not seem unjustified to use low molecular heparin or warfarin, Greenfield filter, or intra jugular stent, especially in the patients with predisposing factors such as hypercoagulability or vasculopathy.[31,32]

Pain should not be treated with nonsteroidal anti-inflammatory drugs. These block the production of prostalglandins, thus attenuating tumor necrosis factor inhibition, resulting paucity of clinical symptoms and the onset of shock and multi organ failure. Adjuncts to therapy may include IV immunoglobulin, activated protein C, and hyperbaric oxygen in order to improve the immune response against the bacterial super-antigens. However, further controlled studies are warranted to support these suppositions.[33,34]

In his original series of 20 cases, Lemierre reported the survival of 2 patients.[1] In the postantibiotic era mortality rates range from 4% to 12%.[6]

According to Riordan, a substantial number of LS cases were reported in the first half of the 20th century. Consequently, publications of LS between 1955 and the 1980s seem to have become scarce.[7] An explanation for this decrease may be the extensive use of antibiotics in treating oro-pharyngeal infections in that time frame.

Many authors agree that there is a resurgence of LS in the last 30 years.[19,28] This fluctuation matches a more judicial use of antibiotics than in the past. Another possible explanation for this resurgence is antibiotic resistance in general.[35]

CONCLUSION

LS is a rare condition. About 4% of all LS cases are of odontogenic origin. In this review, we have shown that LS due to odontogenic infection may be at least as virulent as nonodontogenic LS. Our evidence implies that the causative micro-organism is not confined to the traditional *F. necrophorum*. It seems that other species such as alpha hemolytic streptococci are involved.

The once "forgotten disease" is on the rise with mortality 5–10%. Successful outcomes rely on acquaintance with the syndrome together with vigilance of the clinician to allow for prompt surgical and medical treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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