

# A case of descending necrotizing mediastinitis and Lemierre's syndrome associated with an odontogenic infection

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## ABSTRACT

**Introduction:** Descending necrotizing mediastinitis is a rare complication of odontogenic infections, with a high mortality rate despite the advent of antibiotics. **Case Report:** An otherwise well 37-year-old male presented with descending necrotizing mediastinitis secondary to a mandibular second molar infection. He required prolonged targeted antibiotic treatment, extensive oral, neck and thoracic surgery. After a 68-day intensive care unit admission he was stepped-down to the ward for further monitoring. This admission was complicated septic shock, Lemierre's syndrome and a right phrenic nerve injury. **Conclusion:** Odontogenic infections are common presentations to the local dentist yet many patients delay definitive management, placing them at increased risk of severe complications. This clinical case highlights the multiple potentially fatal complications (including Lemierre's Syndrome) associated with descending necrotizing mediastinitis originating from an infected mandibular second molar.

**Keywords:** Dentists, Lemierre's syndrome, Mediastinitis, Sepsis, Toothache

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## INTRODUCTION

The management of odontogenic infections is primarily entrusted to the dental practitioner. It is imperative that dentists are well versed in potential local and systemic complications associated with odontogenic infection as fatal complications still occur, despite the advent of antibiotics. Odontogenic infections are the most frequent cause of descending necrotizing mediastinitis (DNM), a rare and aggressive necrotizing fasciitis typically originating from an oropharyngeal source and tracking along fascial planes of least resistance into the mediastinum [1]. The DNM has mortality as high as 40% despite aggressive antimicrobial, surgical and supportive care [2]. This review presents a complicated case of severe DNM secondary to an untreated odontogenic infection.

## CASE REPORT

A 37-year-old male presented to the emergency department with a five-day history of lower right-sided mandibular pain, intermittent pyrexia, chest pain and malaise. He had a prolonged history of recurrent toothaches, having been advised by his dentist that he

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required definitive management for grossly decayed molars. Prior to presentation he experienced rapid development of right-sided neck swelling, extreme pain radiating down into and throughout his thorax, severe diaphoresis/dyspnea/odynophagia/trismus, a hoarse voice and the inability to tolerate lying flat. The patient regularly smoked illicit drugs (i.e., marijuana, crystal methamphetamine) with an otherwise non-contributory medical history.

On initial examination the patient was afebrile at 37.0°C and hemodynamically stable with a blood pressure of 142/76 mmHg. He had grossly carious right sided maxillary and mandibular second molar teeth and a 15 mm interincisal opening. There was swelling in the right submental and submandibular regions with erythema and swelling extending down to the base of the neck. He was exquisitely tender to palpation throughout his neck and over the anterior chest wall. His blood tests revealed elevated inflammatory markers with a white cell count of  $27.9 \times 10^9/L$  (normal range =  $3.7-9.5 \times 10^9/L$ ) and C-reactive protein of 475mg/L (normal range  $\leq 3\text{mg/L}$ ).

Computed tomography (CT) imaging revealed extensive bilateral soft tissue and subcutaneous emphysema in the submental, submandibular, parapharyngeal and prevertebral regions and in the carotid spaces. Further subcutaneous emphysema was seen to extend into the anterior and posterior mediastinum as well as the right axilla (Figure 1).

Empirical broad-spectrum intravenous (IV) antibiotics were provided in the emergency department (benzylpenicillin, metronidazole and ceftriaxone) prior to transfer to the operating room. Emergency surgery was immediately undertaken where an awake fiberoptic endotracheal intubation was performed. The carious teeth were extracted along with exploration and washout of purulent material from the submandibular and submental spaces. Septic shock requiring vasopressor support developed during the early postoperative period. At this point he returned to theatre for bilateral anterior thoracotomies and sternal division (clamshell thoracotomy) and an anterior cervical fasciotomy. Necrosis was present in the pretracheal strap muscles, right carotid sheath as well as bilateral necrosis throughout anterior mediastinum (with purulent effusions) requiring extensive debridement and the placement of multiple cervical, intercostal and mediastinal drains (Figure 2).

Vasopressors were weaned in the days following surgery and his antibiotic therapy was extended to meropenem and vancomycin to cover the suspected polymicrobial infection. Mediastinal and cervical tissue cultures grew *Streptococcus anginosus* and a heavy growth of mixed anaerobes, in particular *Fusobacterium* spp. This necessitated targeted antibiotic therapy with a short course of clindamycin and long-term benzylpenicillin.

In the context of continued sepsis progress CT imaging revealed multiple parapharyngeal, retropharyngeal, supraclavicular and right common carotid artery fluid

collections. There were pleural effusions, a pericardial effusion, abdominal free fluid and a right internal jugular vein (IJV) thrombus (Lemierre's disease), 70–90% stenosis of superior vena cava (SVC) and inferior vena cava (IVC) distension. A therapeutic heparin infusion was commenced for management of his venous thrombosis.

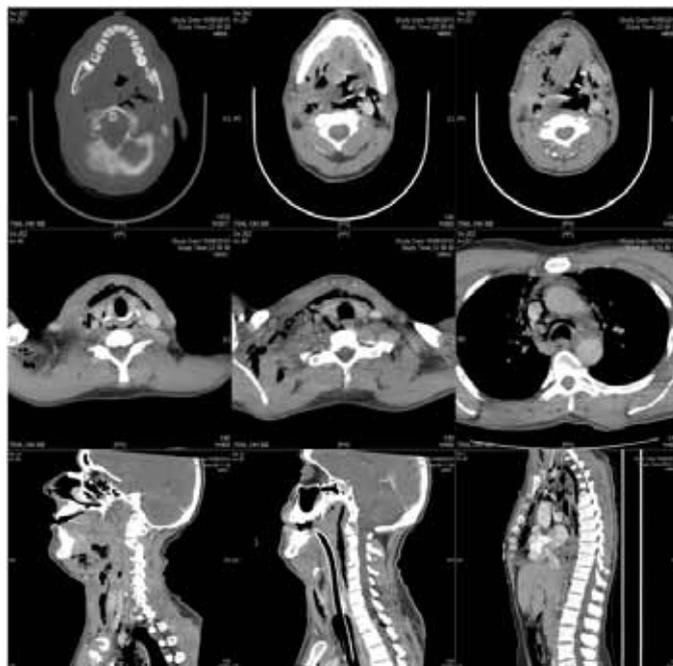


Figure 1: Contrast computed tomography scan. Coronal and sagittal cuts displaying a preoperative carious right mandibular second molar, extensive gas locules demonstrated within the parapharyngeal, pre-vertebral, submandibular and submental regions, the subcutaneous fat of the neck and carotid spaces. This extends into both the anterior and posterior mediastinum as well as the right axilla.



Figure 2: Chest radiograph, displaying an intubated patient with multiple intercostal and mediastinal drains post clamshell thoracotomy.

Multiple repeat washouts with debridement were required due to recurrent collections (Table 1). A notable finding was necrotic tissue surrounding the phrenic nerves bilaterally, with evidence of necrosis along the right phrenic nerve.

The patient failed multiple ventilator weaning attempts and required placement of a tracheostomy due to prolonged intubation. He remained on high levels of pressure support ventilation owing to profound critical illness myopathy along with a possible unilateral right phrenic nerve injury. His antibiotics were later changed back to Meropenem, to cover for an *Enterobacter aerogenes* pneumonia associated with purulent bronchial secretions and persistent pyrexia. There was a staged withdrawal of the intercostal and mediastinal catheters, with no further surgical treatment required.

The patient was nutritionally supported with total parenteral nutrition though the acute phase of his admission, suffering significant feeding intolerance on commencement of an enteral feeding regime. He then required a nasojunal tube for feeding and a nasogastric tube for gastric decompression until the gastroparesis abated.

Table 1: Timeline of operative management

Day	Surgical Procedure	Team
1	Removal of 47 + 48 + intraoral washout + debridement	OMFS
1	Cervical fasciotomy + washout + debridement	OHNS
1	Bilateral anterior thoracotomies + sternal division + mediastinal debridement	CTS
3	Repeat Mediastinal washout + debridement	CTS
6	Repeat cervical washout + debridement + wound closure	OHNS
6	Gastroscopy assessment of oesophageal wall integrity	UGI
6	Repeat mediastinal washout + debridement + closure of clamshell wound	CTS
6	Repeat intraoral washout + debridement	OMFS
12	Repeat intraoral washout + debridement	OMFS
12	Tracheostomy + repeat cervical exploration + washout + debridement	OHNS
16	Repeat cervical exploration + washout + debridement	OHNS
16	Repeat intraoral washout + debridement	OMFS

Abbreviations: OMFS Oral and Maxillofacial Surgery, OHNS Otolaryngology Head and Neck Surgery, CTS Cardiothoracic Surgery, UGI Upper Gastrointestinal Surgery.

Progress CT imaging showed progressive diminution of infective collections. He was continued on long-term antibiotics and was gradually weaned off mechanical ventilation and transferred to the ward after a 68-day intensive care admission.

Whilst on the ward the patient required intense optimisation from allied health teams to facilitate discharge home three months post admission with regular community and outpatient follow-up.

## DISCUSSION

Descending necrotizing mediastinitis is a rare disease most frequently seen in young men during the third to fifth decade [3]. The term descending necrotizing mediastinitis was first proposed by Estrera et al. [2], when describing mediastinitis complicating oropharyngeal infections. A meta-analysis by Corsten et al. [3], reported an overall mortality of 31% and mortality as low as 19% in those receiving both neck and thoracic drainage. Estrera et al. [2], established a diagnostic criteria requiring: clinical manifestations of severe oropharyngeal infections; radiological evidence of mediastinitis; intraoperative or post-mortem verification of necrotizing mediastinitis; a causal relationship between the oropharyngeal infection and mediastinitis.

The necrotizing infection can originate from varied cervicofacial locations, most commonly it complicates odontogenic infections (58%) followed closely by retropharyngeal and peritonsillar/tonsillar infections [4, 5]. In this case the grossly carious lower second molar was clearly the infective focus with copious purulent exudate in the adjacent submandibular space.

An appreciation of deep cervical fascia is imperative to understanding the path of infection and in facilitating safe surgical management. In our patient, the initial CT scan revealed involvement of the submandibular space (adjacent to the odontogenic infection). There were further gaseous locules extending throughout the pharyngomaxillary, anterior visceral, prevertebral and visceral vascular space, providing access to both the anterior and posterior mediastinum.

A high index of suspicion is required for early diagnosis of those with suspected deep neck space infections. Delay in diagnosis is one of the main reasons for the high mortality [4, 6]. Contrast enhanced CT imaging is the most appropriate imaging modality to provide an early diagnosis, at a point where plain film imaging may appear normal [3, 6].

The microbial flora of DNM is typically polymicrobial with both aerobic and anaerobic bacteria cultured in approximately 75% of cases [4]. Neck and mediastinal cultures from our patient grew *Streptococcus anginosus* and *Fusobacterium* spp. with a heavy growth of mixed anaerobes. These oral commensals are in keeping with the typical polymicrobial growth of DNM.

Septic shock is the most common complication of

DNM. It has been reported to develop in as many as 1:2 patients [7]. This patient's illness was complicated by sepsis, recurrent cervical and mediastinal collections, bilateral pleural effusions and pneumonia, a pericardial effusion, right IJV and cephalic vein thrombus (Lemierre's Syndrome), carotid sheath erosion, phrenic nerve injury, gastroparesis and profound critical illness polyomyopathy. Lemierre's syndrome is a rare and potentially fatal septic thrombophlebitis of the internal jugular vein or tributary complicating oropharyngeal infections, acting as a source of septic emboli [8]. The most frequently isolated pathogen is *Fusobacterium necrophorum*, consistent with the *Fusobacterium* spp. cultured in the above patient [9]. Treatment of Lemierre's syndrome involves IV antibiotics and removal of the infective source. The use of anticoagulation is controversial yet in our case a heparin infusion was commenced followed by long-term subcutaneous low molecular weight heparin, to good effect [8, 9].

Multidisciplinary management of DNM involves initial resuscitation, commencement of empirical antibiotics and protecting the airway. Source control is mandatory with debridement and drainage of infected regions.

This case highlights the importance of timely management of all odontogenic infections. In our case, the patient was aware that he required treatment for three carious molar teeth. He delayed this treatment, not being aware of the potentially lethal sequela. Patients need to be aware that delayed treatment of infected teeth not only compromises the dentist ability to provide conservative care but prolongs the exposure to an infective source that may develop into a rapidly fatal mediastinal infection.

Strategies aimed at reducing the incidence of untreated odontogenic infections will have beneficial flow on effects. Medical professionals should be reminded that early surgical management of odontogenic infections by dentists is mandatory and isolated use of antibiotics may contribute to worse outcomes [10].

Dentists should have a low threshold for immediate referral of any patient with signs of deep neck space infections. Concerning signs and symptoms when associated with odontogenic infections include; odynophagia, neck swelling, dysphagia, dyspnea, dysphonia, subcutaneous crepitation and chest pain.

## CONCLUSION

Descending necrotizing mediastinitis is a rapidly fatal complication of odontogenic infections. Dentists should aim to remove the source of any odontogenic infection in a timely manner and all patients who delay treatment should be informed of the dire sequela that are known to complicate odontogenic infections.

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## Author Contributions

Jared Drew Weston – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

## Guarantor

The corresponding author is the guarantor of submission.

## Conflict of Interest

Authors declare no conflict of interest.

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